DYSFUNCTION IN THE FAMILIES OF ANOREXIA NERVOSA AND CYSTIC FIBROSIS PATIENTS

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This thesis has been composed by myself. Apart from the initial interviews with anorexia nervosa and cystic fibrosis patients (carried out by Mrs Wilma Warwick as described in the test), the work is my own.

Caroline Blair
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ABSTRACT

Since the late nineteenth century clinicians have been claiming that the families of Anorexia Nervosa (AN) patients are strange. Some have argued that the families' behaviour serves either to cause or maintain the illness. Most previous work in this field has been confined to self report measures or in the case of interview and observational studies has either lacked a control group or had a comparison group of well families only. It is possible that unusual behaviour could be the natural reaction of any parent to the presence of a major illness in one of their adolescent children.

The present study compared the families of 27 Anorexia Nervosa patients with the families of 29 Cystic Fibrosis (CF) patients and a third group of 31 well controls. The individual child in each household was aged between 14 and 24. Assessment was carried out at baseline and at an eight month follow up. Data was collected via three modalities: (1) Self report questionnaires (2) the Camberwell Family Interview which was rated for Expressed Emotion (EE) and (3) Observation of a family problem solving task which was rated for evidence of Minuchin's "psychosomatic family" dimensions.

The self report measures of family functioning for the most part did not distinguish between the three groups. There were no differences between groups in levels of EE criticism. AN and CF households showed more emotional over-involvement (EOI) than did the well households and EOI correlated with severity of illness suggesting that this family characteristic is an illness related phenomenon. Mothers of both CF and AN patients showed more emotional disturbance than did mothers of well young people. There were more enmeshed and overprotective households in the AN group than in the CF and well groups. AN families were also less successful problem solvers than the other two groups giving partial support to Minuchin's conceptualization. The robust psychological health of CF patients found in previous research was replicated here and was in marked contrast to the AN sufferers. No conclusive relationship was found between change in health status and change in household climate between baseline and follow up. This could in part be explained by design limitations. AN families were more likely to feel that they had been blamed for their child's illness than were CF

families. A brief qualitative account of the AN and CF families in the study showed that they had had very different experiences of getting appropriate support.

A supplementary project was carried out with 214 teenagers and their families in 2 secondary schools. The objective of the study was to develop a self report scale (Edinburgh Family Scale) that could measure Minuchin's "psychosomatic family" concepts for use in the main study. In addition, Scottish normative data was collected on the Family Assessment Measure and on a life events schedule for teenagers.

INTRODUCTION

Anorexia nervosa (AN) is a baffling illness of unknown aetiology, difficult to treat and with a poor prognosis. Many of the treatments in use today were in use 30 years ago and the drop in mortality rate is debatable and dubiously attributable to current therapeutic practices (Hsu, 1987; Treasure, 1991).

For at least 200 years clinicians have been claiming that the families of anorexia nervosa patients are bizarre. The late nineteenth century physicians who named and epitomised the syndrome observed an intense involvement of families with the patient and considered this involvement to hinder recovery. In the past decade both clinical and research interest in families has accelerated since the publication of work by 2 clinical teams: the Milan group (Selvini-Palazzoli, 1978) and the Philadelphia group (Minuchin et al, 1978). Both groups espoused a family system viewpoint and stressed the importance of the family in the development and maintenance of AN. Both claimed outstanding results for their own brands of family therapy. Neither group has tested their theories or therapies with controlled trials. Nevertheless, their work has led to a widespread enthusiasm for family therapy as a treatment method for AN.

Minuchin claimed that the families of AN patients typically showed a particular constellation of family characteristics. He claimed that they were rigid in their structure, enmeshed in their communication, overprotective and incapable of resolving conflict and that these characteristics served to maintain the illness.

Disturbances have been found in the families of many psychiatric patients and also in the families of children with physical illnesses where there is no suggestion that the illness has a psychogenic cause. Do AN families in some way cause or maintain the illness or is their behaviour a reaction to illness, the natural response of any parents to the presence and burden of

a major illness in one of their adolescent children? It has yet to be demonstrated that AN families are more uniquely strange than other families coping with illness.

A recent well designed study (Russell et al, 1987) found that family therapy was more effective than individual therapy in maintaining weight gain in younger, less chronic anorexic patients but did not report on family process changes. This finding has posed several interesting questions. What are the family characteristics that might predict a poor prognosis? There are suggestions from the Expressed Emotion research that negative affective style could be a significant factor. Does this apply equally to the families of physically ill children?

The present study compared the families of AN patients with those of cystic fibrosis (CF) and a third group of well families. The families were followed up 9 months after the initial assessment. The first objective of the study was to investigate whether any of the characteristics thought to be indicators of family dysfunction were present in the families of AN patients more frequently than in the families of a young person whose illness has a clear cut non-psychogenic origin. The second objective was to address the conundrum of whether disturbed families maintain AN or whether their behaviour is a response to illness. What happens to families over time? If dysfunction is a response to illness crisis, then a correlation over time between severity of illness and severity of disturbance would be predicted in both CF and AN families. If a correlation was found only in the AN families, then Minuchin's hypothesis that AN families maintain illness is fostered.

Why cystic fibrosis? The reasons why CF was selected as the comparison group are discussed in Chapter 5.1. Suffice it to say here that it is an illness with a clear cut non-psychogenic aetiology that has a major impact on family life with much parental involvement in treatment. Dietary complications and low body weight after puberty are often a feature.

Chapter 1 is a brief overview of the relationship between physical illness, individual psychopathology and family functioning. Chapters 2 and 3 review the literature on these relationships as they pertain to AN and CF. Chapter 4 reviews the literature on one of the main measures of family functioning used in the study: Expressed Emotion. Chapters 5 and 6 describe the methodology and results of the baseline study. Chapter 7 describes the procedure and results of an 8 month follow up of the participants. In chapter 8 is a qualitative account of the experiences of some of the families in the study. A supplementary project was carried out on 214 adolescents and their families the objective of which was to develop a questionnaire used in the study and to obtain Scottish normative data on 2 further questionnaires. This is covered in chapter 9. In chapter 10 is the discussion. The appendices include copies of the less familiar measures used in the project.

CHAPTER 1 LITERATURE REVIEW - FAMILIES AND ILLNESS

1.1 The Well Adjusted Family

Before launching into a review of families and illness, it would be prudent to pause to consider what is meant by a well adjusted family. Few researchers have attempted to grasp this nettle (Walsh, 1982). One possible explanation is that it is too tedious or obvious to do so. Commentaries on family life often unfold with Tolstoy's much quoted opening lines to Anna Karenina "All happy families are alike but an unhappy family is unhappy after its own fashion" and pass quickly on to the unhappy ones. But happy families are not all alike. Even in Anna Karenina, key families are both happy and unhappy at different times in their life cycle. In the novel the sources of both happiness and unhappiness are different for different families: domesticity, sexual desire, philosophical enquiry and country living are variously both rewarding and punishing to different individuals and families.

A second possibility is that this neglect is a matter of expediency. Only disturbed families or vulnerable families cross the threshold of clinicians and are therefore convenient subjects of enquiry.

A third possibility is that well adjusted families do not exist at all. Much of the work on families focuses on negative events and dysfunctional patterns of interaction. Where results do suggest that families are doing well, in coping with illness, for example, it is not unknown for the finding to be disregarded with the suggestion that "denial" is at work or that underlying pathology is masked (Turk and Kerns, 1985).

There is a commonly held view that disruption to family functioning is a normal consequence of adolescence. Rutter and colleagues (1976) challenged this assumption in a well considered review of the field. To give a flavour of this prejudice, they give some pertinent quotations:

> "Adolescence is by its nature an interruption of peaceful growth ... the upholding of a steady equilibrium during the adolescent process is in itself abnormal ... the adolescent manifestations come close to symptom formation of the neurotic, psychotic or dissocial order and merge almost imperceptibly into ... almost all mental illnesses."
>
> Freud A, 1952

> "Personally, I would feel greater concern for the adolescent who causes no trouble and feels no disturbance."
>
> Geleerd, 1957

Adelson (1964) has argued that much clinical opinion on the adolescent stage is informed by 2 caricatures: what he calls the "victimizer", ie the delinquent leather jacketed amoral character and the "visionary victim", the sensitive, intense, estranged middle class adolescent on whom the psychoanalytic theory of adolescence is almost exclusively based.

Epidemiological studies of adolescence have not in the main supported this negative view of adolescence (Epperson, 1964; Douvan and Adelson, 1966; Graham and Rutter, 1973). For example, the Douvan and Adelson (1966) study, a questionnaire survey of 3000 American adolescents, found that, of all other people, parents were most admired, and one quarter claimed never to have had major arguments with their parents. On the other hand, many had disagreements about clothes, dating and time keeping and about four fifths reported that they would like their parents to be less restrictive. Normal adolescents shared with their parents a common core of values in spite of sharp disagreements about modes of dress, hair style, music and how late they can stay out (Rutter et al, 1976).

A more recent review (Offer and Schonert-Reicht, 1992) adopts much the same thesis as the Rutter et al (1976) review arguing against 5 myths of adolescence: (1) that normal adolescent development is a time of turmoil (2) that adolescence is a time of increased emotion (3) that puberty is a negative event for adolescents (4) that adolescence is a time of increased risk for suicide (5) that adolescent thought is childlike.

However, Rutter and colleagues (1976) did find that half of the healthy 14-15 year olds they studied in a community survey had some inner turmoil (not amounting to psychiatric illness), most of which went unnoticed by adults. 20% of their sample said that they felt that what happened to them was less important than what happened to other people.

On balance it seems likely that well families of adolescents do exist and there have been several attempts to define the dimensions along which healthy families can be distinguished from dysfunctional ones. The factors which particular therapists will regard as important will depend to some extent upon that therapist's orientation. An integration of the various points of view has not yet occurred but attempts have been made (eg Barnhill, 1979 and Fleck, 1980).

1.2 Families and Illness

Much of the relationship between the family and illness remains closed to public view. Many investigations have found that a person defines his or her symptoms by consulting family members (eg Vincent, 1963; Suchman, 1965; Picken and Ireland, 1969). Research studies indicate that 70-90% of all sickness episodes are handled outside the formal health care system, and self treatment within the family provides a considerable proportion of health care. Even when professional help is sought and therapy prescribed, families still retain the responsibility for making decisions about the management of therapies within the confines of the home. Mothers appear to evaluate symptoms of their children by relating them to previous symptoms of other family members (Turk and Kerns, 1985). The family remains the primary unit of health care.

However, there now exists an extensive literature on the relationship between the family and illness (Shapiro, 1983) although little is known about the influences of the extended family or indeed other social networks on illness. Three dimensions of this will be briefly examined here: the

relationship between individual psychopathology and family dysfunction, between individual psychopathology and physical illness and between family dysfunction and physical illness. Correlational evidence for all 3 associations exist. What is much less clear is the direction of causation although much research appears to proceed as if it was known.

1.3 Individual Psychopathology and Chronic Physical Illness

There is now a considerable body of evidence establishing the higher prevalence of psychiatric disorder or distress in persons with chronic medical conditions. This is true for all age groups although research on adults and children has proceeded quite separately, limited perhaps by some discontinuity between adulthood and childhood in the 2 main classification systems for psychiatric disorder, DSM and ICD.

1.3.1 Childhood Disorders

A major review of research in the previous 10 to 15 years on the risk that childhood chronic physical disorder confers for maladjustment was that published by Nolan and Pless (1986). The authors ruthlessly discarded weak designs: case reports and inadequately controlled studies with small sample sizes, and focused mainly on prevalence surveys, cohort follow up studies and on the handful of experimental or quasi-experimental designs available. The bulk of the evidence suggests that children with chronic disorders have twice the risk for secondary emotional handicaps.

Nolan and Pless cite 3 large prevalence surveys, the Monroe County, Rochester (USA) study (Pless and Roghman, 1971), the Isle of Wight (UK) studies (Rutter et al, 1976), and the Michigan (USA) study (Walker et al, 1981) which all come to much the same conclusion. Since the Nolan and Pless review, Cadman and colleagues have reported on the Ontario Child Health survey (Cadman et al, 1987; 1991). Using a stratified household

sample of 3294 children aged between 4 and 16, the age and sex adjusted odds ratio for one or more psychiatric disorders for those with chronic illness alone was 2.1. For those with chronic illness and disability, the odds ratio was 3.4. Psychiatric disorder was broken down into neurosis, conduct disorder and attention deficit disorder (ADDS) using DSM-III criteria. The results from this survey point to large increases in risk for neurotic disorder, ADDS and social isolation for those with disability. A further recent survey carried out in Monroe County, New York, this time of children attending primary care paediatricians (Weiland et al, 1992) found raised prevalence of both emotional problems and behavioural problems in children with serious chronic disorder.

It is prudent to remember that in all these studies the majority of children with chronic disease do not have problems. In Cadman's survey 60% of children with chronic disease involving considerable limitations on usual childhood activities had no psychiatric problems.

1.3.2 Adult Disorders

There is also a large literature on the relationship between psychopathology and physical illness in adults. Most of this work takes place in separate enclaves such as "cancer and depression" or "heart disease and anxiety" with little reference to other physical or psychiatric diagnoses although the tasks of assessment, problem definition and even treatment are often the same. As mentioned above there is also little allusion to childhood disorders in the adult literature. While the psychiatric conditions of childhood falling under the general rubric of "conduct disorders" and their relationship to physical illness is adequately documented, there is relatively little research on the "conduct disorders" of adulthood: the personality disorders, antisocial or criminal behaviour and physical illness. This I suspect is partly because at adulthood many conduct disorder patients leave the psychiatric services and become clients of social work and prison

services, and partly because of the as yet unresolved difficulties in defining and describing the shifting sands of personality disorder (Tyrer, 1988).

A rare study linking childhood and adult disorders was that carried out by Rodgers (1990) who looked at a national birth cohort followed for 36 years to compare the life chances of individuals with chronic physical illness in childhood with those of controls. Women who had been chronically sick as children were more likely to have psychiatric symptoms (PSE) at age 36.

The few studies that provide a broad overview and comparison of psychological status in patients with different chronic illnesses have found a high prevalence of psychiatric disorder or distress in people with chronic medical disease. For example, a large community study from the NIMH epidemiologic catchment area programme (sample size 2554) compared 8 chronic medical disorders for the prevalence of psychiatric DSM-III criteria illnesses (Wells et al, 1988). They found a 41% increase in the relative risk of having any recent psychiatric disorder as a function of having a chronic medical disease. Affective, anxiety and substance use disorders were each more prevalent in persons with chronic medical conditions.

Much research has focused on the relationship between chronic illness and depression. Katon and Sullivan (1990) review the literature on depression and cancer, neurological illnesses, cardiac disease, rheumatoid arthritis, diabetes, and end stage renal disease and find an increased prevalence. Anxiety and physical illness has received much less attention. The Wells study cited above (1988) found that anxiety disorders were particularly prevalent in persons with current arthritis and heart disease.

Although most of correlational work between physical illness and individual psychopathology implicitly assumes that illness causes personal distress, there is some work looking at the effect individual behaviour has on illness. Most of this centres on the concept of compliance with treatment. As an example of this Masland (1982) found that the major cause of low blood

plasma levels with anticonvulsant medication, medication which manages to control seizures in 80% of epileptic patients, was due to failure to take pills in the first place and that psychological problems in turn led to poor compliance.

1.3.3 Risk Indicators for Psychosocial Dysfunction

Now that the link between emotional health and chronic illness is well established, there has been a more concerted search for more specific attributes that further define the risk among subgroups of the population.

The relationship between severity of illness and risk of maladjustment is complex. The problem is exacerbated by the absence of measures of severity of illness that can cross illness boundaries (Stein et al, 1987). Within some disease groups there is uncertainty over how severity should be measured. A further problem is that parental assessment of severity, often mediated by stress factors in the parents, may reach different conclusions from clinicians' ratings of severity (Perrin et al, 1989). Maternal reports of child behaviour are likely to yield as much about mother-child interaction as about the child's behaviour (Lancaster et al, 1989).

The 2 large scale studies by Stein and Jessop (1984b) and Cadman and colleagues (1987) cited above both found no relationship between psychosocial disorder and traditional medical morbidity measures such as bed days. Heller et al (1985) assessed children with congenital heart disease, cleft lip and palate and hearing impairment and found that disease severity was directly related to both the persistence and onset of maladjustment. Severity of illness seems an important factor in juvenile rheumatoid arthritis (Daniels et al, 1987; Ungerer et al, 1988). Summarising data from a series of good prevalence surveys, all of which used structured psychiatric interview as the measurement tool, Katon and Sullivan (1990) showed a linear increase in the prevalence of major

depressive disorder when comparing community, primary care and inpatient adult medical populations from 3% to 6% to 11%.

A review (Pless and Nolan, 1991) of those chronic illnesses in which disorder visibility is prominent found that psychosocial outcome was better when the disorder could be seen. Perplexingly the opposite conclusion was arrived at by Eiser's (1990) review.

There is now considerable evidence that for children cerebral involvement in the diagnostic entity is an important predictor of emotional problems (Rutter, 1981; Breslau and Marshall, 1985; Breslau, 1985; Pumareiga et al, 1990; Weiland et al, 1992). For example Weiland's (1992) study of children looked after by primary care paediatricians found that children with serious disorders involving the CNS to be at particularly high risk for both behavioural and emotional disorder.

There are indications that age affects adjustment. The risk of emotional problems in the general population increases with age (Rutter and Quinton, 1984) and the same is true for children with chronic physical disorder (Pless et al, 1972). Younger children seem more affected in terms of school tasks and achievement (Allen and Zigler, 1986), older children in terms of social adjustment (Ungerer et al, 1988). However, in adults older age groups seem to show less maladjustment (Cassileth et al, 1984). Factors which could be important but have not been researched are the age of onset and duration of the illness as a proportion of life span.

Finally a dysfunctional family places the chronically sick person at risk of emotional disturbance. Using various self report instruments of family functioning, several studies have found that family dysfunction is associated with emotional problems among children with chronic illness (Pless et al, 1972; Pless and Satterwaite, 1973; Friedrich, 1979; Sabbeth, 1984; Kovacs et al, 1985). The relationship between individual psychopathology generally and family dysfunction will be discussed more fully in section 1.5.

1.4 Families and Chronic Physical Illness

A number of different research strategies have attended research on families and physical illness. Much of the work focuses on attempts to measure family burden or the impact of the illness on the family as a whole or on individual family members. Another strand has been thought to measure "family pathology" or family dysfunction as a whole. Finally family coping skills have been looked at.

As with the literature on individual psychopathology and illness, it is difficult to make broad statements about families and illness because research tends to progress in enclaves of particular illnesses with little reference to other disease literature. For example, there are now extensive literatures on families and diabetes (Hauser et al, 1986; Johnson, 1988; Coyne and Anderson, 1988), families and asthma (Gustafsson et al, 1987) families and epilepsy (Kerns and Curley, 1985), traumatic brain injury (Kerns and Curley, 1985), and families and cancer (Kellerman, 1991; Brown et al, 1992). The insularity of these research groups is unfortunate because interesting lines of research followed in one field could be pursed in other disease entities.

An interesting model that has been harnessed as a way of viewing the relationship between families and illness is family systems theory. A brief account of family systems theory will be given in the description on Minuchin's "psychosomatic family" given in section 2.5.3.

1.4.1 Impact on the Family

Traditionally research concerned with the impact of illness or impairment on the family has been based on the proposition that illness or impairment in a family member has severe effects on family functioning. The families of ill people were seen as generally functioning more poorly than families in which all the members are healthy. With the onset of an illness, the

family's social life contracted to become primarily family centred. Within this circumscribed existence the patient often became the focus of the family, with other family members forced into the background. Furthermore most researchers took the view that the more severe and long lasting the illness or impairment, the greater the potential for family disruption. The assumption was that the experience was associated with marital disruption and distress or psychopathology in individual family members. Much of the early work took place on small clinic samples and case studies with no control groups and was heavily dependent on reports from the female caregivers (for reviews see Lavigne and Burns, 1981; Turks and Kerns, 1985; Eiser, 1985, 1990).

However, the large scale community study by Cadman and colleagues described above (1991) found no increased risk for family dysfunction in families with a chronically sick child. Family dysfunction was measured using the General Functioning Scale of the Family Assessment Device (Epstein et al, 1983). This confirmed the findings of Pless and Satterwaite (1973) in an earlier community study.

Reviews of marital adjustment have concluded that the parents of chronically ill children are no more likely to divorce than others (Sabbeth and Leventhal, 1984; Perrin and Maclean, 1988). However, mothers in particular are more likely to have raised levels of distress than in well families. The Ontario study (Cadman et al, 1991) found that the odds ratio for parental treatment for "nerves" was 2.1 in the case of mothers and 1.9 for fathers. They also found increased maternal negative affect scores, confirming observations and findings from many smaller scale projects.

A number of studies point to maladjustment in siblings of chronically sick children (for reviews see Drotar and Crawford, 1985; Lobato et al, 1987). There are suggestions that siblings have lower self concepts, can be socially isolated and resentful of parents' involvement with the sick child. The focus of much of this work has been on identifying the potentially negative

aspects of illness. However, most of this work has taken place on referred samples. There has been some movement away from this deficit centred perspective to look at the supports that siblings can give to disabled people (Eiser, 1990).

1.4.2 Risk Factors for Family Dysfunction

It is likely that the impact of an illness on a family differs in intensity depending on the disease (eg its nature, time since onset, degree of disability involved, degree of stigma attached), several patient variables (eg gender, age and identity of the sick person) and many family related variables, such as overall health of various family members, socio-economic status, the age and stage of the family life cycle at which the illness occurred and the status and position of the sufferer within the family constellation. Cancer in an 80 year old has a different meaning for the family than the same disease in an 18 year old. Moreover, there is some evidence that some families may function more appropriately when there is a sick family member, as if mobilised to more effective functioning (eg Brown et al, 1982; Spaulding and Morgan, 1986).

1.4.3 Coping with Illness

Much is known about the prevalence of maladjustment but relatively little about coping resources within the family. It is also a field which is remarkably thin on theory. There is a body of work on families and coping strategies (reviewed by Shapiro, 1983) but by and large coping and stress models remain at the descriptive level and it is hard to find a theory from which falsifiable hypotheses can be generalised. However, recent work emphasises the enormous range of coping resources displayed by families in relation to both practical and emotional difficulties. There is movement towards viewing individuals with chronic disease as normal people coping with specific stressors rather than as a pathology prone group (Kazak, 1989; Perrin and Maclean, 1988).

1.5 Individual Psychopathology and Family Discord

The best risk marker for most mental disorders is the rather crude index of being the off-spring of a parent with mental disorder (Goldstein, 1988). That there is an association between psychiatric disorder in parents and children is now well established. The earliest reports were careful clinical descriptions of the phenomenon (eg Janet, 1925). The association has been demonstrated in numerous epidemiological studies of the general population (eg Buck and Laughton, 1959; Hare and Shaw, 1965; Richman et al, 1982; Rutter and Quinton, 1984). Further information regarding this risk has come from retrospective studies of patients with psychiatric disorders, studies of children whose parents are being treated for psychiatric disorders and studies of child psychiatric populations.

Can any general statements be made about the association between families and individual psychopathology? Firstly, the link might reflect genetic transmission. There is evidence for genetic factors in schizophrenia (Gottesman and Shields, 1976), in major affective disorder (Gershon, 1984; McGuffin et al, 1991), in antisocial personality disorder and adult criminally (Crowe, 1983) and in some varieties of alcoholism (Bohman et al, 1981; Cloninger, 1981). Genetic factors are probably less important in the wide spectrum of depressed and anxious patients seen in out-patient clinics and community settings. In this population Goldberg and Huxley (1992) suggest that genetic factors could determine a generalised vulnerability towards affective illness, possibly related to one's degree of emotional reactivity and that differing syndromes of minor disorder are partly determined by factors learned within the family and partly by differing environmental factors occurring later in life.

Secondly, there is evidence that mental disorder may interfere with parenting functions. A 4 year prospective study of newly referred psychiatric patients found that children in these families had a substantially increased risk of developing psychiatric disorder during

childhood. An ongoing follow-up of the same children until they reach adulthood found that the main risk did not stem from the illness per se but from associated psychosocial disturbance in the family, particularly personality disorder of anti-social type, parental hostility, irritability, violence and marital disorder (Rutter and Quinton, 1984; Quinton et al, 1990). Parker has argued that a parental style of low care (low warmth) and overprotection (restrictiveness) are implicated for virtually all psychiatric conditions (Parker, 1983, 1989).

The experiential precursors of depression have been the subject of extensive work by Brown and colleagues (Brown et al, 1986; Bifulco et al, 1987; Andrews et al, 1990; Harris et al, 1986, 1990). Lack of care, maternal apathy, neglect and physical and sexual abuse have all been child rearing factors found by these authors to predict later depression. Parental loss increases risk of psychiatric disorder and delinquency in early adult life (Rutter, 1985) but appears to pre-dispose to depression only if it leads to inadequate child care (Birtchnell, 1988; Parker, 1983; Harris et al, 1990). The crucial factor seems to be the quality of care, in particular neglect. The quality of maternal care can be impaired by depression (Weissman, 1972). Chronically depressed mothers have been shown to be less involved with their children and less likely to be empathic or to interact with them in a positive way (Cox and Mills, 1983; Radke-Yarrow et al, 1988).

Third, there is evidence for the burden that psychiatric illness places on families (see Fadden et al, 1987 for review); the loss of reciprocity of relationships, the breakdown of normal communication, the decline of family problem solving skills, the reduction or loss of normal sexual behaviour, upset in the normal pattern of affection and cohesion. These are in addition to the hardships outlined in the section on family burden and physical illness. Particularly difficult to bear are behaviours caused by illness when they overlap with ordinary behaviour patterns. For example loss of interest, loss of communication skills, excessive dieting, lack of interest in self care. Orford (1987), in his study of the families of

profoundly mentally handicapped children, found that mothers were able to tolerate almost any amount of physical burden but were distressed by psychological disturbance: unpredictable behaviour and aggressive behaviour.

This chapter has been no more than a skate over the complex relationships between families and illness but is the context into which research on anorexia nervosa families and cystic fibrosis families must fall and it is to this literature that I now turn.

1.6 Summary

1. Disturbed family life is not a necessary condition of adolescence. (1.1)

2. Chronic physical illness confers twice the risk of psychiatric disturbance in children. However, most chronically ill are not disturbed. (1.3.1)

3. In adults chronic physical illness confers greater risk of depression, anxiety and substance abuse. (1.3.2)

4. A tentative conclusion is that severity of physical illness is a risk factor for psychological disturbance in adults but not in children. Evidence for illness visibility as a risk factor is equivocal. (1.3.3)

5. Cerebral involvement in the diagnostic entity is an important predictor of psychological disturbance in children. (1.3.3)

6. Dysfunctional family life places the chronically sick person at risk of emotional disturbance. (1.3.3)

7. While clinic studies suggest that chronic illness confers risk of family dysfunction, this has not been found in community studies. Marriages are no more likely to break up in parents of a chronically ill child than in parents of well children. (1.4.1)

8. Mothers of physically ill children run twice the risk of suffering from "nerves" or mental ill health than do mothers of well children. Fathers are unaffected. (1.4.1)

9. Recent research has tended to focus on family coping skills rather than family dysfunction. (1.4.3)

10. There is a well established association between psychiatric disorder in parents and in children. Genetic factors play a part in some illnesses but are probably less important in the wide spectrum of depressed and anxious patients seen in outpatient departments. (1.5.1)

11. Mental disorder may interfere with parenting functions. Parenting styles of hostility, violence, irritability, lack of care, maternal apathy, neglect, physical and sexual abuse are risk factors for disturbance in their off-spring. (1.5.1)

12. There is evidence for the burden that psychiatric illness places on families. (1.5.1)

CHAPTER 2: LITERATURE REVIEW - ANOREXIA NERVOSA

2.1 Clinical Features

2.1.1 Weight and Eating Factors

The cardinal feature of anorexia nervosa is the relentless pursuit of thinness which is most commonly expressed by wilful starvation and excessive exercising and less often by vomiting, chewing and spitting, and laxative abuse. This is associated with a fear of weight gain or fatness and a fear of loss of control. The effect of food restriction is severe emaciation. A drop in body weight to at least 85% of expected weight for age and height is the present diagnostic criterion (DSM-III-R) for weight loss. Malnutrition can have profound consequences for the patient including amenorrhoea, ovarian abnormalities, osteoporosis, electrolyte disturbance and cardiovascular complications. If weight loss is not reversed, death will ensue.

Many features of AN behaviour are common to all weight and diet conscious individuals: rigidly held views on acceptable upper weight limits, lists of allowed and not allowed foods, daily weighing, preoccupation with regional fatness particularly stomach and thighs. What is not common to all dieters is the passion and intensity of these preoccupations. Tears and rages can follow the attempts of family to "interfere" or when they have knowingly or unknowingly infringed the "rules" about eating.

Much AN behaviour is a consequence of starvation. A famous experimental study that observed the effects of semi-starvation on a group of American World War II (Keys et al, 1950) conscientious objectors recorded behaviour commonly found in anorexics: a preoccupation with food and eating to the exclusion of all other interests, ritualistic behaviour associated with eating such as only eating at a fixed time, lengthy preparation of meals, meals that can drag on for hours, arrangement of food on the plate in fixed

patterns. AN patients will sometimes insist that no one enter the kitchen while they are preparing a meal or alternatively will only eat a meal if prepared by one person (usually mother). They will often only eat when alone.

Behaviours not seen in starving people are buying large quantities of high calorie food and hoarding this food, and cooking large meals which are then pressed on to reluctant family members but not touched herself. While not wishing to stray into the arena of aetiology, these behaviours are probably best understood as a way of testing self control to the limit, generating a heady excitement over resistance of temptation.

The Department of Social Security defines a "household" as a unit in which housekeeping, cooking and eating arrangements are shared. When these arrangements break down, as they do in anorexia nervosa, the impact strikes at the heart of the household. Ann Erichsen (1985), the mother of an AN patient, gives a graphic account of a family meal.

> "Vegetables are cut into tiny pieces, carefully arranged and later eaten at a maddeningly slow pace. The mother who is cooking for everyone else at a normal speed is driven to distraction by this. She becomes increasingly impatient, eventually exploding with anger ..... By the time the meal is ready, the atmosphere is appalling. Efforts at conversation dwindle, tensions increase. The father, egged on by the mother's martyred expression, loses his temper only to be rounded on by the mother with forceful remarks about his ineptitude. The patient throws a fit and her plate, rushing out of the room leaving behind a boiling pot of bitterness, frustration, guilt and anger."

2.1.2 Exercising Factors

AN patients display a general restlessness and lack of concentration, sequelae of the starvation process. Many also take part in exercise regimes that go way beyond the level recommended for general fitness. This may include behaviour such as standing in the house instead of sitting down

with the family, getting up very early to do work outs with or without exercise machines that may last several hours, "working off" meals by going out for walks of several miles, doing isometric exercises in bed, running up and down stairs unnecessarily.

2.1.3 General Psychopathology

There is universal acceptance that AN patients have a high level of general psychopathology or neurotic symptoms. This has been demonstrated in numerous dimensional studies (eg Stonehill and Crisp, 1977) where general psychiatric symptomatology was closer to that of psychiatric outpatients than well control subjects. It has also been demonstrated in categorical studies where co-morbidity with other psychiatric disorders has been assessed. Studies using case histories, parental interviews, self report questionnaires and structured interviews have shown a high lifetime prevalence of affective disorder (Cantwell et al, 1977; Hendren, 1983; Gershon et al, 1984; Toner et al, 1988; Halmi et al, 1991) and anxiety disorder, particularly social phobia and obsessive compulsive disorder (Rivinus et al, 1984; Toner et al, 1988; Halmi et al, 1991). Evidence for raised levels of alcoholism remain equivocal (Halmi et al, 1991).

2.1.4 Personality Factors

Patients with AN have been described as shy, neurotic, introverted, obsessional, overcontrolled, dependent and perfectionistic (Dally, 1969; Bruch, 1974; Strober et al, 1985; Leon et al, 1985).

> "There is ample evidence from objective study and clinical observation that certain personality traits cluster with unusual prominence in anorexic individuals. These include persistence, a tendency towards rigidity, methodical problem solving, emotional restraint ..... obsessive worrying and poor adaptability to change."
>
> Strober 1991

Parents often describe their daughter as having undergone a personality change with the onset of the illness. Heightened obsessiveness was observed as a feature of starvation (Keys et al, 1950). Unlike Key's subjects, with AN patients this characteristic does not always disappear on weight restoration although it may improve (Garner and Garfinkel, 1982).

A meaningful discussion of the personalities of AN patients is hampered by the lack of any consensus on how to measure "personality" which inevitably is an abstraction inferred from observed behaviour. While it is evident from the few available studies that the **overall** diagnosis of personality disorder has satisfactory reliability and reasonable predictive validity (Robins, 1966; Merikangas and Weissman, 1986; Tyrer, 1988), evidence for stability or reliability of trait defined subtypes is poor (Mann et al, 1981; Tyrer et al, 1988).

There has been a flurry of research in recent years into personality characteristics of eating disordered patients largely due to a rather uncritical enthusiasm for the DSM-III and DSM-III-R personality disorder classification system and the several structured interview schedules and self report questionnaires that have emerged to serve it. These "DSM" studies suggest that personality disorders are present in most eating disorder patients, the most common diagnoses being made in the "anxious-fearful" cluster and the "dramatic-erratic" cluster (Piran et al, 1988; Gartner et al, 1989; Wonderlich et al, 1990; Steiger et al, 1991). Attempts have been made to divide AN subjects who binge "bingers" and those who do not "restricters" on the basis of personality type but no consistent pattern has emerged (Garner et al, 1985; Piran et al, 1988; Gartner et al, 1989; Wonderlich et al, 1990; Steiger et al, 1991). Kennedy et al (1990) urge caution in interpreting DSM studies on personality after finding that a high proportion of their AN patients received widely differing personality diagnoses when assessed on admission and again at discharge.

The high percentage of AN patients deemed to have personality disorders is probably an accurate reflection of the average clinician finding these patients "difficult".

> "Personality disorder has traditionally been a diagnosis with rather pejorative overtones and a diagnosis of despair. When a difficult patient has been assessed and treated fully without a favourable response, the problem is often re-formulated as a diagnosis of primary personality disorder."
>
> Tyrer et al, 1991

2.1.5 Self Esteem

Low self esteem is so commonly found in psychiatric patients that it is sometimes used as an indication of psychopathology (Ingham et al, 1986). Researchers have repeatedly demonstrated that there is a significant correlation between self-esteem or self concept and satisfaction with body characteristics or physical attraction and that the relationship is stronger for the female than the male (Lerner and Moore, 1974; Gray, 1977; Tobin-Richards et al, 1983). Furthermore, in women more than men, physical attractiveness is related to how an individual is evaluated by her peers and to her own personal prestige (Lerner, 1969; Staffieri, 1967). Therefore, it is no surprise to find that low self esteem is commonly found in AN patients (Eckert et al, 1982) and that this is present independent of depressed mood (Silverstone, 1990).

2.1.6 Social Functioning

Social functioning is a commonly used indicator of psychological functioning. As with other starving people, the anorexic gradually narrows her interests. Many entirely restrict their activities to exercise, schoolwork and dieting while all other occupations fall by the wayside. Many girls lose interest in their friends early in the disorder. The social pressures to eat from family and friends also hasten withdrawal from the social activities involving eating and drinking, ranging from refusal to join family meals to

never entering a café or pub. The isolation results in loneliness and a sense of social inadequacy. Restricting anorexic patients tend to be more socially isolated than bulimic-anorexics (Garfinkel and Garner, 1982).

2.1.7 Psychosexual Functioning

AN patients usually lose sexual interest and avoid contact with the opposite sex. In part this represents a complication of starvation but frequently persists after weight restoration. Lack of sexual interest is a common feature of most psychiatric illness (Garfinkel and Garner, 1982; Scott, 1987).

Crisp et al (1980) reported that 25% of their patient series avoided sexual contact before the onset of illness. Without a control group the significance of their finding is difficult to assess. Rothschild et al (1991), using a self report inventory, found that his inpatient sample of eating disordered patients scored at below the first percentile of a sexual functioning index and that 50% of the sample described their present sexual relationship as poor. In a long term (20 years) follow up of AN patients, only half of the group had married and had children (Ratnasuriya et al, 1991). Bulimic anorexics tend to be more sexually active than restricting anorexics but do not describe their relationships as pleasant (Garfinkel and Garner, 1982).

2.1.8 Education and Work

While it is often alleged that AN patients shine as schoolchildren and students, their actual qualifications are no more than would be expected from their social class (Crisp et al, 1980). Treatment for AN with long spells of inpatient or day hospital attendance will by definition necessitate a disruption to education and work. Anecdotal evidence suggests that working capacity is relatively less impaired in AN than the ability to have social and sexual relations or to live independently from parents Hsu

(1980). Reviewing prognostic studies, Steinhausen et al (1991) found two thirds of former AN patients had normal work or educational careers.

2.1.9 Autonomy

There have been marked sociological changes in the West in patterns of leaving home. Goldscheider and Le Bourdais (1986) report that the proportion of 26 year old women who were still living with parents dropped from 37% to 13% between the 1930s and the 1960s. Therefore judgments of "normality" need to be interpreted in this context.

There are many anecdotal accounts of AN patients having difficulty in separating from the family of origin and leaving home (Dally, 1969; Bruch, 1978; Crisp, 1980. For example, Ratnasuriya et al (1991) describe a patient twenty years after onset of illness who telephoned her mother every day for support. How common such behaviours are in the population at large is unknown. Newson and Newson (1963) in their classic study of patterns of childrearing in Nottingham reported considerable numbers of young mothers who spent every day at the home of their mothers only returning home when their husbands were due back from work. This presumably was normal behaviour in working class Nottingham.

2.2 Epidemiology and Natural History

There are wide discrepancies in reported incidence and prevalence in AN, probably due to methodological artefacts rather than geographical or chronological variations (Treasure, 1991). Data from hospital records is likely to underestimate true incidence. A community study in the Netherlands in which a stratified sample of family doctors registered morbidity data using DSM-III-R criteria reported an incidence rate of 6.3 per year per 100,000 population. Only 63% of these patients had been referred to specialised mental health care. The point prevalence was 18.4 per 100,000 population. Of this group 82% were receiving mental health

care (Hoek, 1991). As AN is an illness of adolescence and young adulthood, a number of studies have looked at prevalence in restricted age bands. The best of these is the Swedish study (Rastam et al, 1989) which looked at the entire population of Göteborg 15 year olds, traced through schools and clinics, and found a lifetime prevalence in children of aged 15 and under to be 400 per 100,000, 0.7 per 100 in the case of girls. Extrapolating from Hoek's (1991) figures, there are probably about 900 people in Scotland currently suffering from AN.

In the Netherlands population study described above (Hoek, 1991), 42% of new cases of AN were in the 15 to 19 age group, 21% in the 10-14 age group and 21% in the 20-24 age group. These figures reflect the findings of most series on age of onset. Mean duration of illness in Ratnasuriya et al's (1991) study was 3.7 years. In a community study of London schoolgirls, Patton (1989) found that 41% of girls diagnosed as AN cases had improved spontaneously 12 months later. A recent review suggested that half AN patients have a good outcome, 30% an intermediate outcome and 20% a poor outcome (Steinhausen et al, 1991). Another recent review cites a combined morbidity and mortality rate of 25% in studies of general referral patients (ie not specialist teaching hospitals) (Treasure, 1991). In the short term the standardised mortality ratio for AN is 6 times greater than expected (Patton, 1988). In studies carried out in the 1980s mean mortality rate was 4.4%. However, after 20 years or more of illness, the mortality for chronic populations is as high as 18% (Halmi et al, 1975; Theander, 1985; Ratnasuriya et al, 1990; Steinhausen et al, 1991). Suicide is as common a cause of death as malnutrition.

The distinction between anorexia nervosa and bulimia nervosa (BN) is by no means clear cut. Some 50% of normal weight bulimics (in clinic populations) give a definite history of AN. Families studies show family clustering of AN, BN and subclinical AN in relatives of AN patients, suggesting that these disorders may represent variable expressions of a common underlying psychopathology (Hsu, 1990).

Some years ago Dally (1969) suggested that no recovery was possible after 7 years of illness. However, patients are reported to be still recovering after 12 years (Theander, 1985; Ratnasuriya et al, 1991).

2.3 Aetiology

The prevailing view is that causes of the illness are multi-dimensional.

> "While all theories acknowledge the role of earlier experience in helping to set the background for the development of the disorder, those which especially emphasise early events postulate specific developmental sequences. Formulations based on issues that appear later in psychological development emphasise the role of current attitudes, beliefs and relationships in bringing about and maintaining the disorder. These latter accounts view anorexia nervosa as a common solution to a more varied set of problems."
>
> Garner and Isaacs, 1985

The main factors which have been postulated as playing a predisposing role in the genesis of the disorder are listed below. The theories receive only brief mention here and will receive more attention later only in so far as they involve families.

2.3.1 AN as Genetic Disorder

Holland et al (1988) suggests that 80% of the variance in liability to AN may be accounted for by genetic factors (Treasure, 1992).

2.3.2 Neurophysiological Factors

Kaplan and Woodside (1987) reviewed recent investigations into the hypothalamic pituitary axis, the central nervous system amines and peptides, carbohydrate metabolism and gastrointestinal hormones but came up with no convincing evidence for predisposing factors although it seems possible that biological mechanisms may play a part in maintaining illness.

Many of the hormonal and neuro-endocrinological abnormalities noted in anorexia nervosa are thought to be secondary to the effects of starvation.

2.3.3 Anorexia Nervosa as Atypical Depression or OCD

While there is an association between depressive illness and anorexia nervosa established through epidemiology studies, family history and genetic studies (Swift et al, 1986), the prevailing view is that there is more divergence than overlap between the 2 disorders, arguing against a common aetiology (Strober and Katz, 1987; Strober et al, 1990). Rothenberg (1990) argues for AN as being a special form of obsessive compulsive disorder but the aetiology of that illness remains obscure too.

2.3.4 Psychoanalytic Models

Numerous formulations are found here (reviewed by Goodsitt, 1985) ranging from drive-conflict postulations that self starvation is "a defense against sexual fantasies or oral impregnation" to object relations hypotheses (Selvini-Palazzoli, 1974; Masterson, 1977).

2.3.5 Autonomy and Identity Factors

The term "ego identity" was introduced by Erik Erikson (1955) to describe "both the persistent sameness with oneself and a persistent sharing of some kind of essential character with others". Hilde Bruch (1974, 1978) has been the most influential exponent of anorexia nervosa sufferers having failed to meet the challenge of adolescence and adulthood. She looked for the causes of AN in the developmental history of the child. She viewed anorexia nervosa as developing amid 3 related ego disturbances - distortion of body image, distortion of internal perception and a sense of personal ineffectiveness. She postulated that the search for self mastery and autonomy was maladaptively pursued through control of the body. However, as she observed herself, problems with autonomy and separation

are not unique to anorexia but are a common feature of adolescence (Rutter et al, 1976; Hsu, 1989; Offer and Schonert-Reichl, 1992).

Conceptually linked to Bruch's position is the Garner and Bemis model that taps into cognitive aspects of ego identity (1982, 1985). These authors claim that anorexia nervosa is in part (at least) caused and maintained by faulty assumptions and irrational beliefs about selfhood as well as about shape and weight. They and Slade (1982) have argued that positive reinforcement such as feelings of success, being in control and self satisfaction is magnified in a person who perceives the rest of her existence as a failure.

Historically, fasting has been associated with religious observance rather than a desire for slimness. There are clear parallels between the search for religious virtue, sacrifice, penitence, asceticism and the desire for specialness and personal effectiveness in the AN patient (Bell, 1988).

2.3.6 Fears of Psychosexual Maturity

Crisp (1980) has argued for many years that AN is an attempt to cope with intense fear of psychobiological maturity. Dieting and "weight phobia" have the effect of a regression to a prepubertal state. The regression is reinforced by the relief it provides from adolescent turmoil and related conflicts within the family.

2.3.7 The Addiction Model

This model is more concerned with how AN is maintained than how it is caused. The theory developed by Szmukler and Tantam (1984) postulated that once begun AN became self perpetuated through addiction. There has been more subsequent interest in the theory a propos of BN than of AN (Vandereycken, 1990)

2.3.8 Socio-cultural Theory

The culturally desirable attractive female form in the West is a slim one. Women desire a thin body and there is some evidence that the desire is intensifying (Garner et al, 1980). At the same time, young men and women in the West are becoming heavier (Christensen et al, 1981). Dubow et al (1990) found that 53% of adolescent girls compared to 16% of adolescent boys were concerned about their weight. Several studies have shown a relationship between the degree of acculturation to Western ideals and eating disorders: in multi-cultural schools in Zimbabwe (Hooper and Garner, 1986), in Kenyan Asian immigrants to England (Furnham and Alibhai, 1983) and in Americans of Hispanic origin (Pumareiga, 1986). Increased body weight in the West is associated with lower socio-economic status (eg Halmi et al, 1978; Braddon et al, 1986; Garn and Clark, 1975, 1976). Adolescent females at every level of fatness in the higher social classes want to be thin more often than those in lower classes (Dornbusch et al, 1984). The fact that anorexia nervosa is more common in the sector of the population that is most commonly engaged in dieting and lays special emphasis on a slim body (women, the upper social classes, and in Westernised populations) suggests that culturally determined dieting and desire for slimness is a powerful precipitant of an eating disorder.

2.3.9 Family Aetiology Theory

Several authors have argued from different theoretical standpoints that the family is of aetiological significance in anorexia nervosa (eg Bruch, 1974, 1987; Selvini-Palazzoli, 1974; Minuchin et al, 1978; Crisp, 1980). This literature will be reviewed in section 2.5.

2.4 Treatment

If there are multiple theories of AN, there are also numerous treatments. Treatments tend to cluster in 2 areas: those that focus on weight restoration per se and those that focus on the patient's feelings, sensations and ideas.

Weight restoration methods that have some short term efficacy include nursing care with high calorie diet and total or modified bed rest (eg Dally and Sargant, 1960), operant conditioning (eg Garfinkel et al, 1977; Kreipe and Kidder, 1986; Schmidt and Marks, 1989), coercive treatments such as hyperalimentation and tube feeding (eg Maloney and Farrell, 1980) and dietary advice (Hall and Crisp, 1987).

Copious amounts have been written about psychotherapy for anorexia nervosa patients (eg Garner and Garfinkel, 1985) but only a handful of controlled trials have been carried out. There is a some evidence for the efficacy of family therapy (Russell et al, 1987; le Grange et al, 1992b); for cognitive-behavioural and behaviour therapy (Channon et al, 1989); for mixed individual and family therapy packages (Hall and Crisp, 1987; Crisp et al, 1991) and for group therapy (Crisp et al, 199l). Psychoanalysis has not been evaluated in controlled trials nor has the cognitive therapy approach (Garner and Bemis, 1982, 1985).

Kennedy and Goldbloom (1991) reviewed drug therapies for AN and found no role for medication apart from in patients with concurrent psychiatric disorders. There are numerous self help booklets directed both at sufferers or their families (eg Crisp, 1980; Palmer, 1980; Ericksen, 1985) but their efficacy is unevaluated. Most of current research on treatment is carried out by specialist well established treatment centres but little is known other than anecdotally of the treatment of patients managed by their family doctors, of isolated cases on general medical wards or psychiatric wards, by

self help groups, by feminist organisations or by the ever increasing army of lay counsellors.

With such a galaxy of theories and treatments, like astrology, at least some of it works some of the time. Writing in 1980 Kendell made the following biting comment:

> "It [anorexia nervosa] fascinates psychiatrists and several have made it a lifetime interest. Some are professors of psychiatry, all work in teaching centres. But when you look at their research as a whole, the striking omission is that not one of them has attempted to measure the efficacy of the treatment they are advocating."

The major treatment centres tend to hedge their bets and adopt a multi-dimensional approach. A survey of clinicians carried out at the International Conference on Eating Disorders in 1988 and 1990 found that less than 50% of respondents believed there is a consensus on treatment of eating disorders. Talking therapy was overwhelmingly endorsed as a treatment for AN (Herzog et al, 1992).

One of these dimensions, and a central focus of this thesis, is the role of families. Before describing current practice with families, I shall turn to a historical perspective on families and AN.

2.5 The Historical Perspective on Families

2.5.1 The Early Descriptive Accounts - Parentectomy

Lasègue writing in 1873 describes the behaviour of relatives as anorexia nervosa progresses.

> "The family has but two methods at its service which it always exhausts - entreaties and menaces - and which both serve as a touchstone ... the more the solicitude increases, the more the appetite diminishes. The patient disdainfully tastes the new

> viands and, after having shown her willingness, holds herself absolved from any obligation to do more. She is besought as a favour and as a sovereign proof of affection to consent to add even an additional mouthful to what she has taken; but this excess of insistence begets an excess of resistance ... the patient thus gets surrounded by a kind of atmosphere from which there is no escape during the entire day."

A year later Sir William Gull of Guy's Hospital, London, writes

> "The patients should be fed at regular intervals and surrounded by persons who would have moral control over them, relatives and friends being generally the worst attendants"
>
> Gull, 1874

These men of power and influence, soon joined by Charcot (1889) and others, acted on their disapproval of families by advocating hospitalisation and total separation from family members (so called "parentectomy") as the only appropriate method of purging the patient from these noxious influences.

Following the publication in 1914 of a report by Simmonds (a pathologist) of destructive lesions in the pituitary gland of an emaciated woman, the emphasis shifted away from psychological explanations of AN to theories of endocrine disturbance.

2.5.2 The Influence of Psychoanalysis

The ascendance of psychoanalysis in the United States led to a shift in interest in formulation from the syndrome being a rare pituitary cachexia to a neurotic explanation (Lucas, 1981). In drive theories of AN, the illness was conceived of as representing symbolically a repudiation of sexuality or of fantasies of "oral impregnation" (eg Waller et al, 1940; Masserman, 1941) and the shift in interest went from bad parents to bad mothers. This period was the hey day of the "psychopathogenic" mother found too in schizophrenia, asthma and diabetes (Turks and Kerns, 1985). She was at

her most potent in schizophrenia, the "schizophrenic" mother being "overprotective, cold, critical and impervious to her child's needs, thus thwarting his or her ability to differentiate, to form a separate identity and to develop close relationships" (Goldstein and Strachan, 1987).

More recent psychodynamic formulations have de-emphasised drive theory and have focused on early object relations (Selvini-Palazzoli, 1974; Sours, 1980; Goodsitt, 1985). Bruch (1974) and Selvini-Palazzoli (1974) both later abandoned psychoanalysis as a treatment for AN but as a developmental explanation for the onset of the illness invoke the bad parent as a necessary if not sufficient aetiological factor.

Bruch writes in The Golden Cage (1978)

> "The development of AN is so closely related to abnormal patterns of family interaction that successful treatment must always involve resolution of the underlying family problems". "Anorexics grow up in harmonious looking well meaning families where they are often the most valued child, one who at the same time has been the most rigidly controlled" (p106).

Bruch says that failure of the mother to allow the child to make any decisions or gain autonomy leads to the "personal ineffectiveness" that she sees as being the core cognitive dysfunction in AN patients. She also feels that in AN families there is competition between the parents about who is making a greater sacrifice to the child with the concomitant expectation that the child will accede obediently to the parents' extreme demands for approval, good behaviour and success. Selvini-Palazzoli (1974) suggests that the AN mother rewards compliance to her wishes, is over-protective, and is unable to allow separation in the child. Sours emphasises the need for the mother to have a submissive perfect child as her own fulfilment (1974, 1980). The focus of treatment by Bruch and Sours was individual psychotherapy for the child with individual psychotherapy for the mother.

2.5.3 The Influence of Family Systems Theory

At the start of the '70s a new influence on attitudes to AN families started to emerge: the family systems framework. Viewed as a system, families have properties that are more than the sum of their parts, they are made up of subsystems composed of the various members aligned by age, role, and other relationship parameters. Every system and the hierarchies between them are differentiated from each other by boundaries, the properties of which are important in understanding how the family works, particularly how permeable or fixed those boundaries are. The family system tends towards equilibrium or homeostasis. A family problem is best understood with a circular rather than a linear model of causality (Nichols, 1984; Barker, 1986). Eisler (1988) has emphasised that this is a perspective on families rather than a prescriptive account of families.

The 2 most influential advocates of systems thinking in AN have been 2 clinical groups, the "Philadelphia group" (Minuchin, 1967, 1974, 1984; Minuchin et al, 1975, 1978; Liebman et al, 1974a, 1974b) and the "Milan associates" (Selvini-Palazzoli, 1974, 1988, 1989, Boscolo et al, 1987; Selvini-Palazzoli et al, 1989).

It is difficult to sum up briefly the "Milan approach" because since its birth the original 4 members of the team have split up and are working in different directions and some of the main theoretical assumptions have gone through a number of perplexing volte-faces (Selvini-Palazzoli, 1988). Remaining neutral on what constitutes healthy family functioning, the objective of Milan family therapy is to challenge rigid family beliefs so that the family is freed to discover for itself new ways of behaving and relating. During the explorative interview with the family, the therapist is supported by a team of unseen observer co-therapists. During the treatment session the therapist leaves the family to consult with the team to formulate hypotheses about the family and then returns to deliver the team "message" to the family. This message or intervention has 2 parts:

a systemic re-frame when the therapist "re-tells" the family story and a prescription, which is often a behavioural homework task, frequently a somewhat arresting paradoxical injunction.

Selvini-Palazzoli (1974) describes her clinical experiences of treating 12 anorexic out-patients and their families.

> "Behind the facade of respectability and marital unity, the parents generally conceal a deep disillusionment with each other that they are quite unable to acknowledge, let alone resolve".

She characterised these families as having a high degree of marital dysfunction, problems of leadership, a rejection of communicated messages, poor conflict resolution, blame shifting and extreme rigidity. She claimed therapeutic success for all these families but Milan Family Therapy for AN has not been assessed with controlled trials (Carr, 1991).

Minuchin's structural theory was originally developed from his work with Puerto Rican urban slum families in New York (Minuchin et al, 1967). It is set out in a number of publications including the books Families and Family Therapy (Minuchin, 1974) and Psychosomatic Families: Anorexia Nervosa in context (Minuchin et al, 1978). It makes use of many of the ideas derived from systems theory and places particular emphasis on the family structure: the arrangements which govern the transactions between family members. He places particular emphasis on the boundaries between family members and between subsystems in the families and whether these boundaries are enmeshed (when the behaviour of one family member has an immediate and marked effect on others) or disengaged (when stresses effecting one family member have little effect on other family members). He also emphasises the importance of alignments, of the coalitions and alliances within families and the power structure of the family. The aim of therapy is to alter organisational patterns by the therapist joining the family in a strong intervention.

Minuchin's move to Philadelphia in the '70s led to his explanatory model of the structure and functioning of the families of psychosomatically ill children (Minuchin et al, 1975, 1978). This model maintained that 3 factors were necessary for the development of severe psychosomatic illness in children. First the child must be physiologically vulnerable in that a specific organic dysfunction must be present. Second the child's family must have 4 transactional characteristics: enmeshment, overprotection, rigidity and lack of conflict resolution. Minuchin's full description of these characteristics are found in Appendix III. Finally the sick child plays an important role in the family's pattern of conflict avoidance and this role is an important source of reinforcement for his symptoms.

Although Minuchin's ideas about AN were not widely publicised until the '70s, they owe their roots to earlier developments in the field of schizophrenia. Lidz and colleagues (1957) argued that in the families of schizophrenics roles between parents and child were disturbed. Murray Bowen (1978) at about the same time was hospitalising the families of schizophrenics for observation and treatment. He described his families as suffering from

> "undifferentiated family ego mass which is the contagion of feeling within a family so that if one family member experienced an emotion, the emotion reverberates round the family."

Wynne and colleagues (1958) studied patterns of communication in families of schizophrenics and described patterns of rapidly shifting alliances in which no 2 people ever got really close or really angry. They also observed that the boundary of the family appeared to be open but in fact was impervious. He named this the "rubber fence" and explained that in these families there is a strong reluctance to disagree or have differences. These concepts from Lidz, Murray-Bowen and Wynne bear a remarkable resemblance to Minuchin's much later concepts of enmeshment, rigidity and avoidance of conflict resolution.

Minuchin distinguishes 2 types of "psychosomatic disorder". In primary disorder a physiological dysfunction is always present. There is an emotional exacerbation of an already available symptom: the example he cites being asthma and diabetes. The group have been at pains to emphasise that a small sub-group only of children with diabetes should be labelled "psychosomatic" (Rosman and Baker, 1988).

In secondary psychosomatic disorder, no such predisposing physical dysfunction can be demonstrated.

> "The psychosomatic element is apparent in the transformation of emotional conflicts into somatic symptoms."
>
> Minuchin et al, 1978, p.29

The example given is anorexia nervosa. Presumably all AN families are therefore "psychosomatic". The observational study through which the Philadelphia group claim verification for the "psychosomatic family" is described in Section 2.10.3.

With this conceptualisation of the aetiology and maintenance of anorexia nervosa, Minuchin and colleagues placed a clear responsibility on the family to become involved in treatment.

> "Weight gain alone is never considered sufficient. It is only a first step and must be followed by a restructuring of the family system."
>
> Liebman et al, 1983

At the first multi-disciplinary international conference on AN in 1976, Minuchin claimed 88% success rate for his methods. He has always been an enthusiastic publicist of his work through the use of one way screens, demonstrations and videotapes (eg Minuchin, 1982). In these videotapes he is seen to act as a strong, emotionally involved director of the family drama, making flamboyant interventions in the family structure which focus sharply on placing power in the hands of parents. The most

sensational of these interventions is the "family lunch" session when patient, family and therapist eat together. If the patient refuses to eat, parents are given the task of getting her to eat which can involve physical force. If they fail, the therapist underlines their failure as strongly as possible. The aim is to induce a crisis in the family by bringing parents to the point of admitting failure and to redefine ill presenting symptoms by discounting the family's myth that they are fine except for the presence of their sick child. Minuchin has also described and demonstrated intervention techniques for challenging the transactional characteristics he sees as typical of "psychosomatic families" (Minuchin, 1974, 1978).

Until 1987 (Russell et al, 1987) there were no controlled trials testing the efficacy of family therapy for AN yet family therapy approaches to AN, particularly for younger patients, were adopted with uncritical enthusiasm. In the history of psychotherapy 3 magic ingredients have led to enthusiastic adoption of new techniques: charismatic leadership with virtuoso performances of technique, astute clinical observations immediately familiar to experienced therapists, and claims of stupendous success rates. Both Minuchin's and Selvini-Palazzoli's groups met these criteria.

2.6 Current Management Practice with Families

The major centres tend to differ not so much in the main ingredients of the treatment recipe (all pay some regard to weight restoration, ego needs and social adjustment factors) but there are quite wide differences in the quantity and quality of each ingredient dependent on the general philosophy of that unit. There are also wide differences in beliefs about and management attitudes towards families. The following section is not intended to be a comprehensive review but more to give a flavour of current different approaches.

The Toronto hospital programme (Programme for Eating Disorders, 1990) was awarded the APA Gold Achievement Award for 1990. The unit does

not admit patients who are under 18. In the day patient arm of this unit, all significant persons in the patients' lives are involved from the outset of treatment and are expected to attend initial assessment.

> "Both patient and family are expected to become involved in an intensive treatment program with the mutually agreed goals of increased body weight and decreased binge and purge behaviour."

Parents are expected to attend family therapy meetings and group meetings with psycho-educational objectives (Woodside and Shekter-Wolfson, 1991). In the outpatient arm of the unit, assessment and counselling services are routinely offered to families and to spouses/partners (personal communication).

The University Psychiatric Center in Kortenberg, Belgium, has communicated prolifically over the past 15 years and has given several accounts of its work (Vandereycken and Meermann, 1984; Vandereycken et al, 1989). The Kortenberg unit takes patients from age 16 to 45, a third of whom are married. The director of the unit, Walter Vandereycken, stresses the importance of involving the family of origin (1985). Vandereycken has charted his shift from a position where parents were actively kept away from a strict behavioural regime (a practice abandoned largely for the pragmatic reason that the regime was deeply detested by both patient and parents resulting in high treatment refusal and drop out rate) to an approach whose basic philosophy is "to work with instead of against the family" (Perednia et al, 1989). With outpatients, all parents are given information about the illness and methods of treatment and are then given concrete advice on what to do about meals, cooking etc and are asked to agree with the patient on her contract for weight restoration. No in-patient is admitted without agreement that her family will be involved in treatment (Vandereycken and Norre, 1992) either to receive family counselling sessions focusing on information and support or family therapy. They see the families of patients on the point of admission as being families

in crisis but not necessarily pathological families. A small percentage of very disturbed families receive family therapy which has been described in some detail. The main focus is on the here-and-now situation aiming at stimulating change in the actual problems presented by the family. However, the group state

> "We do not have clear-cut and research based criteria for the use of family therapy in general or for the application of specific strategies or techniques in particular."
>
> Vanderlinden and Vandereycken, 1984

A long established unit that has also frequently described its work is the St George's Hospital Unit, London (Crisp, 1980, 1990, 1991). Crisp has a more negative view of AN families than the Toronto or Belgian groups. He postulates that it is essential to identify the "original maturational problems" that he believes were at one time present. To pursue this dormant developmental psychopathology the patient's family of origin must be interviewed, preferably prior to the patient at the initial assessment meeting. He sees presenting family conflicts and parental "deskilling" as being of secondary importance to the developmental adolescence related difficulties (Crisp et al, 1991). Crisp (1988) states

> "The families of anorectics are often characterized by a variety of anxiety avoidance behaviours, eg anorexia nervosa, obesity, alcoholism, the avoidance elements of depression and conflict avoidance behaviour within the family system, particularly by means of noncommunication."

The St George's Unit has adopted a number of treatment packages, all of which involve the family of origin at some level in either family therapy or parental group meetings. With families treatment usually focuses on the here and now rather than then on the developmental maturational problems that are pursued with the patient alone in individual psychotherapy.

All 3 units described so far all involve the family of origin in treatment even though the patient is in most cases an adult. However, an example of a unit that has minimal involvement of families is the Cullen Centre, Edinburgh. This day and outpatient unit has a cognitive behavioural approach, the central features of which are "to promote feelings of autonomy, self control and self esteem" (Freeman, 1992). There is no expectation that parents will be involved in treatment and parents are not interviewed unless this is expressly wished for by the patient. A parent support group run by parents with a small amount of nursing input has now withered away.

Units for younger sufferers tend, often for historical reasons, to be organised separately from adults, a cut-off age of 16 or 18 being commonplace. Thus the Toronto unit described above has no links with the children's eating disorder unit in the Hospital for Sick Children situated some 300 yards away (personal communication).

The most popular treatment method for younger patients would appear to be family therapy. Writing in 1988 Eisler says

> "It is true that family therapy was originally conceived of as a treatment of families, but most practitioners nowadays would describe what they do as treatment with families remaining more neutral to the question both of the origin and current location of the disturbance."

Describing family therapy with young adolescents in the same department (Institute of Psychiatry, London), Dare has given a graphic account of recent practice (Dare, 1991). This mode of treatment has been tested with controlled trials (Russell et al, 1987).

> "We do not think that symptoms are the outcome of a family's structural qualities, although we do observe that there are patterns of family interaction that render a family powerless in the face of symptoms."
>
> Dare, 1991

He states that the aim of treatment with adolescents is for parents to take control of their child's eating pattern, "to set up a meal regimen such as occurs in a well functioning hospital in-patient unit". After an initial meeting whose objective is anxiety augmentation, the next meeting is usually a family meal which has the objective of compelling the child to eat.

> "The therapist should frequently stand or crouch behind the parents, with his or her hands on their shoulders, urging them to be implacable, powerful and irresistible to their daughter."
>
> Dare, 1991

The second stage of treatment turns from a single minded focus on eating to family talk about problems that pre-occupy most families with teenagers. The final phase focuses on the launching phase with leaving home issues.

Parental control of eating is in diametric opposition to the approach of Vandereycken, for example, where responsibility for eating is taken by the therapist and Freeman where it is taken by the patient. Indeed it is different from Dare's approach with older adolescents and young adults where parents are helped to disengage themselves from an involvement with issues of weight and food (Dare et al, 1990). Dare recognises the paradox of 2 techniques, the exact opposite of each other, one aimed at increasing parental power, the other at withdrawing it, appearing to be equally efficacious but does not explain how this can be reconciled with the Bruch view that a central problem of AN is lack of autonomy, self esteem, personal effectiveness and fear of lack of control caused by excessive parental control.

While there is considerable variation in both the amount and the kind of parental involvement in the units described above, they do, albeit with varying levels of conviction, emphasise the need to work with parents and not to see families as "to blame". How far the idea of the pathological family has been dropped from most clinical practice, however, is unknown.

2.7 The Typical Anorexia Nervosa Family

In a masterly review of family issues in anorexia nervosa, Yager (1982) describes the typical anorexic family of popular mythology.

> "In an upper middle class, highly achievement orientated family, mother, and often others, are constantly vigilant about their weight and they all value slimness and physical exercise. Superficially this is a healthy family but diligent about putting up a congenial facade. Certain unaddressed conflicts between the parents lurk below the surface ... parental stresses and concerns are channelled and deflected towards the children so that the mother becomes excessively involved with them ... the parents inadequately acknowledge the child's individuality so that she develops a fragile self image and feels that there are no real areas for self control. Her poor sense of self and accompanying sense of ineffectiveness are ignored by the parents."

Yager's sketch combines descriptions from Crisp and colleagues (1974, 1980), Russell (1977), Bruch (1978), Minuchin (1978), Selvini-Palazzoli (1974) and Sours (1980) and from popular literature and TV documentaries.

As with many psychiatric syndromes, the initial "classic" descriptions of families were based on relatively small numbers, often with skewed samples and with no comparison group. What is the factual basis for this description?

2.8 Are there Demographic Differences between AN Families and Other Families?

2.8.1 Social Class

A very common finding in many studies has been the over-representation of higher social class families with an anorexic child (eg Fenwick, 1880; Bruch, 1974; Dally, 1979; Szmukler et al, 1986; Gowers and McMahon, 1989). Most of the work has been confined to hospital case registers or retrospective clinic surveys, and thus some of this over-representation may be due to the ability of the middle classes to seek out, utilise, and travel considerable distances to procure the expertise of eating disorder specialists. The magnetic effect of specialist centres to the higher classes is a phenomenon well known in many disorders such as cancer and cystic fibrosis.

There is however evidence for the social class bias from other sources. In a much quoted community study (Crisp et al, 1976) great efforts were made to identify unequivocal cases of AN in a survey of English schoolgirls in private and comprehensive schools in South London. The prevalence in girls over 16 was 1.05% in the middle class private schools and 0.05% in the largely working class state schools. A recent Swedish study found greater prevalence of AN in private schools although it is not clear from their study if private schools have the same social class implications in Sweden as they do in Britain (Rastam et al,1989).

Gowers found the usual social class skew when looking at 100 consecutive cases of AN at the St George's Hospital, London but found that the skew was less marked than in a similar survey carried out 10 years previously at the same hospital (Gowers and McMahon, 1989).

The finding of social class bias is not however universal. Leighton and Millar (1985) working in Glasgow surveyed all the NHS psychiatric facilities in their area and did not find a class bias. However, they suggest that

there may be selective referral of social classes I and II away from NHS psychiatric facilities in the city.

Rastam and Gillberg (1991) using a Swedish classification system with 3 social classes reported 50% of AN patients in the lowest social class which is unusually high. This study warrants particular attention both because of the representativeness of its sample and the existence of a control group matched for age, sex and school. Half the sample of 51 AN cases was recruited from a population screen of the total population of Göteborg 15 year olds (Rastam et al, 1989) by school doctors, nurses and through psychiatric clinics. The other half were later referrals from the same school doctors and nurses.

2.8.2 Parental Age

Many investigations have reported that the parents of AN sufferers are relatively elderly (eg Dally, 1969; Theander, 1970; Bruch, 1974; Hall, 1978). However, none of these studies controlled for social class, delayed parenthood being associated with the middle classes. The Swedish study (Rastam and Gillberg, 1991) found no differences in parental age between AN and well control groups.

2.8.3 Family Size and Birth Order

Patients come from a variety of family sizes and from any birth order although there have been reports of various combinations being significant (see Garner and Garfinkel, 1982 and Vandereycken et al, 1989 for review). A large retrospective case note study of 252 patients (Gower et al, 1983) found no effect and no preponderance of female siblings as reported by Bruch (1974) and Hall (1978). Rastam and Gillberg's study (see above) also found no family size effect.

2.9 Is there more Individual Psychopathology in AN Family Members?

2.9.1 Prevalence of Weight and Eating Problems

Several descriptive reports suggest a higher incidence of eating disorders in family members (eg Theander, 1970; Morgan and Russell, 1975; Crisp et al, 1980). However, none of these used blind analysis of systematically obtained interview data on relatives, precise diagnostic criteria or inclusion of a normal control group for comparison. Using these more rigorous methods but failing to match for social class, Gershon et al (1984) found that the combined rate for anorexia nervosa and bulimia was 6 times higher in first degree female relatives of anorexia patients than in relatives of controls (6% and 1.1% respectively). Strober et al (1990) used similar methodology and found that female relatives of AN patients had a fivefold greater risk of eating disorders compared to female relatives of other psychiatric disorder populations and a ninefold risk compared to female relatives of affective disorder populations. Halmi et al (1991) found an incidence of 4/169 of eating disorders in AN first degree relatives compared to 0/178 in control families in their family study. (These findings say nothing over the nature-nurture issue.) It should be noticed that although the rate of eating disorders is considerably higher in AN families than in well families, the low overall prevalence means that at least 90% of families do **not** have an eating disorder.

A combined twin and family study (Holland et al, 1984, 1988) found 56% of the monozygotic twin pairs and 5% of the 20 dizygotic pairs were concordant for AN and 5% of other female first degree relatives also had an AN history. Twin data are subject to confounding influences. A significant number of Holland's twins were recruited through direct recruitment and voluntary referral, strategies which tend to overselect for MZ twins and concordance. Other methods of determining the genetic component of an illness: genetic markers, twins reared together versus twins apart and

adoption studies have not been carried out. In view of virtual disappearance of at birth adoptions away from family in the West (Tizard, 1991), it is most unlikely that adoption studies can now be carried out on this population.

In view of the wealth of information now available about the socio-cultural aspects of weight, slimness and dieting, a second question arises as to whether AN families are particularly potent bearers of that culture (Garfinkel and Garner, 1982). It has been suggested by many observers that family members of AN patients have an excessive interest in food, weight, shape, eating behaviour and activity (Crisp, 1967; Theander, 1970; Bruch, 1974; Kalucy et al, 1977). However, in well controlled studies this has not been observed (Halmi et al, 1978; Garfinkel et al, 1983). Hall et al (1986) studied the weight histories and attitudes towards weight related matters in 58 mothers of AN patients and 204 mothers of schoolgirls of similar age and class and found no support for the "weight pathology" hypothesis. They looked at attitudes to weight, comparing the AN mothers with Weight Watcher attenders and mothers of schoolgirls and found that the AN mothers (assessed by questionnaire) showed a lower concern about weight related matters than all other groups. The authors suggest that this may reflect a certain amount of denial or minimisation.

2.9.2 General Psychopathology in Parents

The raised prevalence of psychopathology in the relatives of most psychiatric disorders would suggest that this ought to apply to the families of AN patients too. However, there are conflicting reports. Some of the uncertainty may be caused by the different methodologies used.

There are many anecdotal and case history accounts of raised prevalence of psychiatric illness in parents. Dally and Gomez (1979) found 24% of patients had parents who had required psychiatric treatment at some time and concluded that the incidence of neurosis was similar to that found in

groups of other neurotic patients. Again taking psychiatric consultation as the criterion, Morgan and Russell (1975) and Crisp et al (1980) found rates of a mental health history similar to Dally.

Using self report questionnaires, Crisp et al (1974) and Garfinkel et al (1983) found no difference in levels of current psychopathology in parents of AN patients compared to controls.

Family prevalence studies suggest that lifetime rates of depression for AN first degree relatives fall between 7% and 22%, 2 to 3 times the rate for control families (Winokur et al, 1980; Hudson et al, 1983; Gershon et al, 1984; Rivinus et al, 1984; Strober et al, 1982, 1990). This was not confirmed by another recent family study (Halmi et al, 1991) although the authors did find twice the rate of one or more psychiatric diagnoses in their AN families (21% v 11%). Rastam and Gillberg (1991) interviewed mothers for current diagnosis and found no depressed mothers in their control group (0/51) and 4/51 with depression in the AN group. Where fathers have been reported on separately, they are usually indistinguishable from controls (eg Halmi et al, 1991). Biederman et al (1985) and Strober et al (1985) found that increased rates of affective disorder were present only in relatives of a subset of AN patients with depression whereas another study (Gershon et al, 1984) found rates of depression equal in relatives of depressed and non-depressed AN patients. Halmi et al (1991) did not find a difference in levels of depression in mothers of restricting and bulimic anorexics although they did find more anxiety disorders in mothers of bulimic anorexics.

Although we can say with confidence that most studies suggest that affective disorder is more prevalent in AN families than one would expect by chance, not enough adequately designed studies have been carried out to comment on other psychiatric disorders. Once again it is worth noting that **most** AN families do not have a history of psychiatric disorder and therefore being raised in a "sick" household is a minority experience for AN sufferers.

2.9.3 Personality Factors in the Parents

Using "common sense" descriptions of personality, Dally (1969) found a great diversity among the parents of his patients. In 21% of families mothers were forceful and robust while fathers were weak and remote; in 17% mothers were tense, neurotic and forceful and fathers were passive, in 5% a domineering, aggressive quarrelsome father was found, in 4% fathers were "psychopathic, unreliable and inconsistently domineering". Kalucy et al (1977) found marked obsessionality as a dominant lifestyle trait in 29% of fathers and 14% of mothers. Both these studies, and there are others in similar vein, were case series with no control group, no blind interviewers and no information is presented on the anticipated prevalence of these traits in the general population. The only controlled study of parental personality is the Göteborg study (Rastam and Gillberg, 1991). Maternal personality disorder was measured using DSM-III-R diagnoses after interview. The authors found that their well comparison group had a higher rate of Axis II diagnoses than the AN group! Once again, the Göteborg study illustrates that in community surveys much less pathology in families is found than is the case in clinic studies. As the evidence stands, personality diversity would appear to be as great in AN parents as in the population at large.

2.9.4 Marital Discord

Marital discord (open quarrelling and criticism) is well established as predictive of childhood psychiatric disturbance (Rutter and Quinton, 1984). The traditional clinical view is that the marriages of AN parents appear happy but that the appearance is superficial (Bruch, 1978). Measured with the index of broken homes, AN families if anything seem more stable than the general population. Halmi (1974) found an incidence of divorce of 18% in her sample which is considerably lower than in the US population at large. In Britain Heron and Leheup (1984) also reported 18% of divorce. Rastam and Gillberg (1991) in their community study found no difference

in marital harmony between the AN and control groups. Minuchin et al (1975) suggest that the AN patient serves to deflect parental conflict which could explain the negative findings of these studies but this would require some evidence that marriages collapse when the patient recovers.

Crisp et al (1974) reported an increase in self reported psychoneurosis (MHQ) in parents following weight restoration in their AN child. However, mean scores were not significantly different from well controls.

2.9.5 What is the Influence of Early Family Experience on Anorexia Nervosa?

There is a long tradition of interest in the influence of early childhood parenting experiences on later disorder, stemming from developmental psychology and from the psychiatric literature (Rutter and Quinton, 1984; Robins and Rutter, 1990). Unfortunately there are no longitudinal studies of childhood cohorts that permit any meaningful focus on AN families. Following 1000 families would produce possibly 10 AN sufferers at age 16.

Relatively little has been written about the pre-adolescent experiences of AN patients. Most authors confine themselves to descriptions of family behaviour on clinical presentation or hypotheses about events surrounding the move from puberty into adolescence (Crisp, 1980). Probably the most influential voice here has been Hilde Bruch (1974, 1978). She has described the early development of a series of 51 patients (see also Section 1.5.1)

> "well cared for children to whom many advantages and privileges of modern living had been offered ... yet on closer study, it was recognised that encouragement or reinforcement of self expression had been deficient."
>
> Bruch, 1974

She goes on to describe the parents of these patients, adding that the description probably applies to many "success orientated" upper-middle class families (60% of her sample were "upper class"). However, she claims

that it is probable that these traits are more pronounced in the anorexia nervosa family. With no control group, this remains speculative.

There have been various attempts to confirm these clinical descriptions by asking AN patients retrospectively about their early childhood experience. Esparon and Yellowlees (1992) gave the EMBU, a Swedish questionnaire aimed at assessing the experiences of parental childraising practices, to 34 women who had suffered from AN or BN in the past 10 years and to 34 medical and nursing staff volunteers. The AN mothers and fathers were seen as less warm and more rejecting than controls but not more over-protective. Design limitations were that no information is available on the current health status of the individuals and the response rate of the eating disorder group was only 50%

The Parental Bonding Instrument (PBI) (Parker et al, 1979, 1989) aims to measure 2 aspects of parental style over the first 16 years of life. 'Care' reflects the warmth and empathy within the relationship, while 'protection' measures perceived overprotection and control by the parents. Two studies (Palmer et al, 1988 and Calam et al, 1990) found no difference between control subjects and anorexic subjects on the protection dimension for mothers and fathers. In Palmer's study mothers only were perceived as showing less care. Fathers did not differ from the control group. Unfortunately neither group used a psychiatric control group. Other studies (ie not AN subjects) suggest that the perception of "affectionless control", ie low warmth and high protection, is a predictor of general psychopathology (Parker, 1989).

Perhaps the most painstaking of research into the childhood experiences of eating disorder patients is the semi-structured interview study of Schmidt and colleagues at the Institute of Psychiatry, London (1993). Using methodology developed for investigating the childhood experiences of depressed women (Harris et al, 1986), the authors gathered information from 64 AN, 116 BN patients and 23 mixed eating disorders in a lengthy

tape recorded interview. This was later rated by consensus raters for a number of variables including change in family structure, quality of childhood care, childhood sexual abuse and perceived approval. Quality of childcare was measured from assessment of parental indifference, low control (laissez-faire), high control, family discord and both physical and sexual abuse. The authors calculated a composite measure of childhood adversity and found that twice as many BN families had high scores as did AN families. Unfortunately, because of the lack of any control group, it is difficult to comment adequately on the patterns of AN childhood experience. The authors found 23% of the AN sample reported lack of parental care, a composite index of high indifference and low control. This compared with 14% and 11% respectively in 2 normal working class community samples in a quite separate study that used the same methodology (Bifulco et al, 1987; Andrews, 1990). 47% of AN patients reported family discord (ie fighting, quarrelling etc) which seems high but no control information is available. In interpreting these findings, it needs to be borne in mind that the Maudsley hospital as a tertiary referral centre attracts a high proportion of difficult cases. Interestingly, these findings are quite at odds with the "polite" family of popular literature (Sours, 1980) or of Bruch's overprotective overcontrolling parents. Levels of sexual abuse were no higher than those quoted in community studies (Bifulco et al, 1991) and reports of physical violence were very low. As there is no corroborating report from, for example, siblings, whether these descriptions of past experience are "true" is unknown. However in the community study mentioned above (Bifulco et al, 1987), depressed subjects recalled childhood experiences in a similar fashion to their well sisters. Schmidt found no relationship between the duration of illness (split into high and low at the median of 4 years) and level of adversity.

2.10 Current Family Functioning

2.10.1 Self Report Studies

The last few years have seen an explosion in the number of self report studies of family environment and family dysfunction in AN patients. The plethora of reports has served largely to confuse the picture rather than clarify it because of the wide variations in both methodological procedures and the instruments used. Some of the difficulties encountered in evaluating this material are as follows:

1. Different instruments attempt to measure different concepts, some with an attributed theoretical background, some apparently without. For example the FACES III measures 2 orthogonal dimensions of family functioning, adaptability and cohesion, which are key concepts in Olson's Circumplex Model (Olson et al, 1985). The Family Environment Scale (Moos and Moos, 1980) appears to have no theoretical base but seems to relate to the values of a very circumscribed middle American cultural group. Sample questions from this scale are "We often talk about the religious meaning of Christmas, Passover or other holidays", and "Someone in our family plays a musical instrument".

2. Several of the studies have no control group (eg Strober, 1981) or control groups of doubtful provenance such as people recruited from a newspaper advertisement (Stern et al, 1989) or paid psychology students (Wonderlich and Swift, 1990).

3. Some have produced a large number of statistics but have failed to use any of the expected checks for Type I errors (Stern et al, 1989).

4. It is not always clear what kind of subjects are being assessed. The term "bulimic-anorexic" is used variously to describe anorexia nervosa patients with a past history of bulimia, patients who currently meet

diagnostic criteria for both anorexia nervosa and bulimia nervosa and anorexia nervosa patients who also occasionally binge and/or vomit.

5. Some authors report the results of patients only, some of parent(s) and patients separately, some a "family" score usually a mean of the family triad.

With all these obstacles, can anything useful be said about the self report literature? Firstly many of the self report inventories measure dimensions that appear conceptually linked, confirmed by correlational and factor analytical studies (Bloomquist and Harris, 1984; Bloom, 1985; Dickerson and Coyne, 1987; Perosa and Perosa, 1990). Conceptually linked scales that have been used in anorexia nervosa research are the Family Assessment Measure (Skinner, 1983), the Family Assessment Device (Epstein et al, 1983), the FACES III (Olson et al, 1985), the Leuven Family Questionnaire (Kog et al, 1985b), the Family Environment Scale (Moos and Moos, 1980) and the Structural Family Interaction Scale (Perosa et al, 1981).

Only studies with a sample size of at least 10 are reviewed. They are listed in Table 2.1.

TABLE 2.1 FAMILY FUNCTIONING IN ANOREXIA NERVOSA: SELF REPORT STUDIES

Authors	Measures	Subjects
Garfinkel et al (1983)	FAM	AN-R v AN-B patients and mothers
Garner et al (1985)	FAM	AN-B v AN-R v BN patients
Grigg et al (1989)	SASB	AN v C patients
Harding and Lachenmeyer (1986)	SFIS	AN v C patients
Houben (1981)	B-L	AN v C triads
Humphrey (1986a)	FES, FACES	AN v C patients & parents
Humphrey (1986b)	SASB	AN v BN v AN-BN v C patients and parents
Kog et al (1985b)	Leuven	AN v BN v C patients and parents
Kog et al (1989)	Leuven	AN v BN v C patients and parents
Leon et al (1985)	FES	AN v C patients and parents
Steiger et al (1991)	FAD	AN-R v AN-B v BN v C patients
Stern et al (1989)	FES	AN-R v AN-B v BN v C patients and parents
Strober (1981)	FES	AN-R v AN-B parents
Waller et al (1989, 1990)	FAD	AN v BN v C patients and parents
Wonderlich and Swift (1990)	SASB	AN-R v AN-B v BN v C patients

Key

Subjects:	AN	= Anorexia Nervosa
	AN-R	= Restricting AN
	AN-B	= AN with Bulimia
	AN-BN	= Meet both criteria
	B-AN	= Bulimia with history of AN
	BN	= Bulimia Nervosa
	C	= Well Controls
Scales:	FES	= Family Environment Scale (Moos and Moos, 1980)
	FAD	= Family Assessment Device (Epstein et al, 1983)
	FACES	= Family Adaptability and Cohesion Evaluation Scales (Olson et al, 1985)
	SFIS	= Structural Family Interaction Scale (Perosa et al, 1981)
	Leuven	= Leuven Family Questionnaire (Kog et al, 1985b)
	FAM	= Family Assessment Measure (Skinner et al, 1983)
	B-L	= Barrett-Lennard Relationship Inventory (Houben, 1981)
	SASB	= Structured Analysis of Social Behaviour (Humphrey and Benjamin, 1986)

Cohesion Studies

The most robust concept measured is that of **cohesion** which taps the extent to which family members are concerned and committed to the family and the degree to which family members are helpful and supportive of each other. The FACES III cohesion scale, the FES cohesion, the Leuven cohesion scale, the FAM affective involvement scale, the FAD affective involvement scale and 4 of the SFIS subscales all seem to be measuring cohesion.

6/8 studies measuring cohesiveness showed no difference between AN **patients** and the control subjects (Garfinkel et al, 1983; Leon et al, 1985; Kog et al, 1985b; Harding and Lachenmeyer, 1986; Stern et al, 1989; Steiger et al, 1991). The 7th showed lower self report of family cohesion from AN patients (Waller, 1989, 1990). The 8th study (Kog et al, 1989) found more cohesiveness in AN patients. 3/4 studies of the **parents** of AN showed no difference from the control group (Garfinkel et al, 1983; Kog et al, 1985b; Stern et al, 1989). The 4th study (Leon et al, 1985) found less cohesion in AN parents. A 5th study by Waller and colleagues (1990) found less cohesion in mothers but not in fathers.

Conflict Studies

The concept of **conflict** taps the extent to which the family openly quarrels or disagrees. Aspects of this dimension are present in the Conflict subscales of the FES and the Leuven Family Questionnaire, Problem Solving in the FAD and Task Accomplishment on the FAM.

No clear position emerges here. 3/7 of the studies show no difference compared to controls (Leon et al, 1985; Kog et al, 1989; Stern et al, 1989), versus 4/7 where raised conflict was found in the AN patients (Garfinkel et al, 1983; Kog et al, 1985; Waller, 1990; Steiger et al, 1991). Garfinkel et al (1983) found raised conflict in AN mothers, Leon et al (1985) and Kog et

al (1985b) in AN parents compared to Stern et al (1989) and Waller (1989, 1990) who found no differences between parent groups.

Expressiveness Studies

Expressiveness measures the extent to which family members are allowed and encouraged to act openly and to express their feelings directly. The Expressiveness Scale on the FES, the FAM Affective Expression Scale and the FAD Affective Responsiveness Scale seem conceptually related.

3/5 studies found no difference between patients and controls (Garfinkel, 1983; Leon et al, 1985; Waller, 1989, 1990) and 2/5 found less expressiveness in patients (Stern et al, 1989; Steiger et al, 1991). Garfinkel et al (1983) and Leon et al (1985) found AN parents to be less expressive. Waller et al (1989, 1990) found no difference in fathers but less expressiveness in mothers. Stern et al(1989) found no difference for parents.

Adaptability Studies

Finally, the concept of **adaptability**. Clinically this is probably most usefully viewed as a concept with 2 negative poles, rigidity at one end and disorganisation at the other, with health functioning somewhere in the middle. In practice, it is usually measured as a unipolar scale with "chaotic" or "disorganised" at the negative pole or "rigid inflexible" at the negative pole and healthy at the positive pole. The Adaptability Scale of both FACES III and the Leuven, the Behaviour Control Scale of the FAD and the Control Scale on the FAM feature here as do 2 SFIS subscales.

7/8 studies found no difference between AN patients and controls (Garfinkel et al, 1983; Leon et al, 1985; Kog et al, 1985b; Harding and Lachenmeyer, 1986; Kog et al, 1989; Stern et al, 1989; Steiger, 1991). Waller et al (1989, 1990) found low adaptability in AN daughters. 6/6 studies found no

differences between AN parents and controls (Leon et al, 1985; Kog et al, 1985; Stern et al, 1989; Garfinkel et al, 1985; Kog et al, 1989; Waller et al, 1989, 1990).

A less well known self report measure is the Barrett-Lennard Inventory which aims to measure perceptions of the quality of relationships within the family. Houben et al (1989) found no overall difference in attitudes to each other between AN patients and their parents and well controls and their parents. The AN daughters viewed their mothers as being more understanding of their (the daughters') emotions and feelings than in the normal girls' experience.

The Structural Analysis of Social Behavior (SASB) (Benjamin, 1974; Humphrey and Benjamin, 1986; Humphrey, 1986a, 1986b) is a conceptually and mathematically complex model of inter-personal behaviour which claims to be able to describe a full array of systemic, interpersonal and intrapsychic events. Its use does not seem to have spread much beyond the originators of the model (Benjamin and Humphrey), possibly because the concepts are so recondite and the scoring system so elaborate. The methodology consists of a set of self report rating scales and also an observational coding system. Using the self report methodology Humphrey (1986b) reports that her bulimic subjects but not her anorexic subjects had severe difficulties in parental nurturance and empathy.

Grigg and colleagues (1989) using the SASB could find no single family pattern that characterised the families of AN patients. Wonderlich and Swift (1990) (in a study which may have been carried out on the same subjects as Humphrey's 1986b study) using the SASB found that once levels of depression had been controlled for, differences between the eating disorder and normal groups disappeared.

Heinmaa et al (1992) found that self reports by eating disorder patients of poor family functioning (FAM) related to whether they were depressed or

not. Their mothers' perceptions of family functioning were unrelated to the daughters' level of depression. They suggest depression serves to cognitively distort the patient's view of family life. The research design does not allow for comment on whether these depressed patients maintain the negative view of families when mood improves.

2.10.2 Interview Studies

In the Swedish community study (Rastam and Gillberg, 1991) a parent from each AN family and each matched control family was given a detailed interview by a psychiatrist (who was not blind to group). The AN families were described as having more problems (unspecified) than the well families but on the whole were very similar. The authors looked for evidence of Minuchin's "psychosomatic family" characteristics. They found no difference between groups for enmeshment, rigidity and conflict avoidance and found that the control families were more overprotective than the AN families. They also found that the AN families were less likely to deny or cover up problems. Small subgroups could be identified who were characterised by "psychosomatic family" features. Unfortunately, they do not describe the questions or scoring methods used to evaluate the psychosomatic characteristics.

2.10.3 Observational Studies

In order to look at the evidence for the existence of the 4 characteristics hypothesised as being typical of psychosomatic families: enmeshment, overprotection, rigidity and lack of conflict resolution, Minuchin and colleagues carried out the following study. Detailed descriptions of these concepts can be found in Appendix III. 11 AN, 9 psychosomatic diabetic and 10 asthmatic families and 15 families with a child with good medical control of diabetes were studied. Half of these diabetic children had behavioural problems. The families carried out a series of Family Tasks: making up a menu together, discussing a family argument, describing what

pleased and displeased them about other family members, making up stories about family pictures and putting together a puzzle. The Family Diagnostic Interview was designed to assess the child's role in detouring conflict. In this task parents discuss a family problem together observed through a one way screen by the child who later joins them to continue the discussion. The family tasks and diagnostic interview were videotaped and later rated by independent raters for the 4 "psychosomatic" concepts. This rating system has not been published, although available in embryonic form from the Philadelphia Child Guidance Clinic. The guide gives clear examples of dysfunctional behaviour but the scoring system is abstruse. The authors state that their observations support their hypothesis but their results are presented in anecdotal form rather than a more usual tabular or statistical form.

Goldstein and colleagues (1981) observed the videotaped interaction of AN family triads taking part in a problem solving discussion. The main interest in these families was as a control group for the schizophrenics' families they had been studying for some time. They found that only 1 of the 11 anorectic families studied adopted a negative affective style using critical, intrusive or guilt-inducing statements, an interaction style later found to be conceptually linked to the critical comments index of Expressed Emotion (Leff and Vaughn, 1985) which is predictive of a relapse in schizophrenia. Goldstein interpreted this result as indicating that the AN families showed less tolerance for conflict rather than the simpler explanation that they were less negative in style.

Using the observational methodology of the SASB (see Section 2.10.1), Humphrey et al (1986) videotaped problem solving tasks and compared family triads of "bulimic-anorexic" subjects with normal family triads. More negative evaluations of the AN triads than the well triads were made by the observers.

Kog and colleagues (1985a, 1985b, 1987, 1989a, 1989b) at the Eating Disorders Unit, Kortenberg, Belgium set out to test Minuchin's psychosomatic family model with a research design that dealt with some of the methodological problems that flawed Minuchin's original work. 30 eating disorder families were compared with 30 normal control families matched for class, family size and age of patient. 83% of the eating disorder patients met DSM-III criteria for AN. Response rate from the well families approached was only 30%. All family members still living at home were asked to carry out the "decision making task" (the interest task) and the "conflict resolution task" (the disagreement task) and were then asked to complete the Leuven Family Questionnaire.

In the decision making task, the family members were asked to complete individual 12 item questionnaires asking for their 2 favourite and 2 least favourite interests or activities. (Sample question "Which type of movies do you like the most or least? Science fiction/historical/psychological film/thriller/cartoon/ comedy/Western"). Minuchin's concept enmeshment was redefined as one pole on a continuum of intensity of intrafamilial boundaries and labelled "boundaries". It was operationalised by adding up all identical interest choices and all different choices between each family dyad. The overall family score was the difference between the dyad most in agreement and the dyad least in agreement. This produces a rather perplexing score in which the families that agreed the most and those that disagreed the most both ended up being described as "cohesive" whereas families in which some dyads agree and others disagree were called "differentiated".

Using the same task, family members were then asked to say "who likes to do what with whom". If family members consistently chose the same member or members for each interest or activity, the authors described that family as having strong "organisational stability", ie that they were inflexible and rigid. They labelled this concept "stability" and used it as a redefinition of Minuchin's concept rigidity.

While the procedure and scoring are easy to follow and involve no subjective choices on the part of the rater, the ecological validity of the decision making task seems light years away from Minuchin's observational studies and his concepts of rigidity and enmeshment and fails to convince as an adequate operationalisation. Although the authors describe the decision making task as a behavioural task, the methodology is in fact paper and pencil self report.

Much more promising was the "conflict resolution" task which was carried out between the subsystems "parents" and "children". The family was allowed up to 30 minutes for this task. The children and the parents each had to come up with a family problem that they explain and then discuss and negotiate the other subsystem. The 2 problem discussions were videotaped and then rated on a 5 point interval scale from "no conflict" to "conflict escalation" to mean "disagreement".

There are however intrinsic difficulties in interpreting the scores. Although the scale can measure the amount of observed disagreement in families, it tells us little about how successful the family is at problem solving. Scores at both poles of the scale "no problem is formulated or discussed" and a "conflict escalation, the discussion ends without resolving a solution" seem unsatisfactory problem solving strategies. A middle score of 3 "compromise, the 2 subgroups have a different proposal for resolution but they agree upon a compromise solution" seems the best problem solving strategy.

These 3 measures "boundaries, "stability" and "disagreement" and the 3 subscales of the Leuven Family Questionnaire "cohesion", "adaptability" and "conflict" evolved out of earlier work by this research group when they attempted to establish the discriminant and convergent validity of operationalisation of Minuchin's concepts using the multitrait-multimethod correlational approach pioneered by Campbell and Fiske (1959). (Kog et al, 1985a, 1985b, 1987). The authors found convergent validity between their

behavioural and self report measures of conflict (r = .35) and their 2 measures of cohesion (r = -.31) but no correlation between their 2 measures of adaptability (rigidity).

The authors found that in the "conflict resolution task" the AN group showed less disagreement or conflict than the well group. They interpret this as supporting Minuchin's notion that AN families avoid conflict, tension and disagreement. An alternative explanation could be that this group are more successful problem solvers than the well group.

On the 2 other tasks, cohesion and adaptability measured on the decision making task, the author found no differences between groups. This could of course mean that there are no differences but equally could reflect the limitations of the operationalisations as discussed above.

2.11 Treatment Studies

What do we know of AN families from treatment studies? Minuchin's structural family therapy applied to AN patients has already been described (Section 2.5.3). He and his colleagues have not carried out controlled trials but reported on a series of 53 patients (Minuchin et al, 1978). The median age of the sample was 14.5 ranging from 9 to 21. Median weight loss at treatment onset was 30% body weight. 40% had had previous treatment. Although younger patients were always seen with their families, older patients quickly moved to individual sessions and their parents to marital sessions. The authors followed up 80% of the sample for 2 or more years and claim that 86% had made full weight and psychosocial recoveries. An unusual feature of the sample compared to most published studies is the short duration of illness (6 months) prior to treatment onset which might account for their exceptional success rate. Nevertheless, the lack of a control group, the absence of data on height/weight ratios, the lack of independent assessors are profound weaknesses in the design. Furthermore, no assessment of family functioning was made so that no

evaluation can be made on whether changes in family structure did indeed take place.

Since Minuchin's report there have been 3 studies that have assessed the efficacy of family treatment. A fourth is in progress. Hall and Crisp (1987) randomly allocated 30 patients with AN to either 12 weeks of outpatient dietary advice or 12 weeks of a mixed individual psychodynamic and family therapy package. The psychotherapy package focused on the role AN played in the relationship of the subject with her family and others aiming to change those aspects of relationships which tended to stifle the subject's development and maintain the anorexia, particularly over-protectiveness, conflict avoidance, enmeshment and distancing within the family. One year after assessment all subjects and their families were interviewed by an assessor blind to treatment allocation. The dietary advice group were found to show significantly better weight gain while the psychotherapy group showed better sexual and social adjustment. It is difficult, however, to draw any firm conclusions from these results. The 2 treatments were not "pure" in that the psychotherapy group also received 4 sessions from dieticians and the dieticians sometimes saw the family as well as the patient. Furthermore, after the 12 week treatment period, up to half the "dietary" sample received some psychotherapeutic treatment from other agencies. The design did not allow a judgment to be made on how much improvement in the psychotherapy group was due to the individual and how much to the family sessions.

A more complex design from the same research group (Crisp et al, 1991) randomly allocated 90 AN patients to 4 options: (1) in-patient treatment of several months followed by 12 sessions of out-patient psychotherapy. Family therapy was part of the package at both the in and out-patient stage: (2) out-patient individual and family psychotherapy plus separate dietary counselling: (3) out-patient group psychotherapy with separate groups for patients and parents plus dietary counselling: (4) no treatment. This latter group were referred back to their local doctor who received a

detailed assessment report and advice on further management. At 1 year follow up the 2 out-patient options had gained significantly more weight than the in-patient group in spite of having considerably less time and effort spent on them. Members of the "no treatment" group had in fact nearly all had treatment of various kinds elsewhere but did less well than the groups under study. What particular aspects of the different treatment packages were helpful remain unclear. Contact time was not a variable, nor was the mode of treatment: group or individual.

To date there is only one study that assesses the efficacy of family therapy per se as a treatment for eating disorders, the landmark study of Russell and colleagues (1987). This study was well designed with a decent overall sample size (57 AN patients, 23 BN patients), assessors independent of therapists, satisfactory baseline and outcome measures and clearly described interventions. Hopefully the group will report on the longer term results of their work. Following discharge from an in-patient unit which aimed to restore nutritional status, patients were allocated to either individual supportive psychotherapy or family therapy. The overall results were not impressive with only 9% of the BN patients and 23% of the AN patients having a good outcome. There was no difference in the efficacy of family therapy v individual therapy. However, in a subgroup of younger, less chronic patients with AN, 90% of patients receiving family therapy had a good or intermediate outcome compared to only 18% of the patients receiving individual therapy. The impressive results for family therapy for this subgroup need to be replicated as cell size was only 10 for the family group and 11 for the individual group.

The same research group are currently engaged in a trial of 2 different kinds of family work: family therapy versus family counselling. They have already reported on the pilot stage of this project (le Grange et al, 1992a; 1992b) in which no differences were found between treatment methods.

2.12 Families and Prognosis

What influence do families have on prognosis?

> "As with many psychiatric disorders, the sickest patients usually come from the sickest families."
>
> Yager and Strober, 1985

Is this true for anorexia nervosa? In a general review of prognostic studies on AN Steinhausen et al (1991) state that a conflict free parent-child relationship was cited as a good prognostic sign in 7 studies and that it was not significant in 2 studies. Unfortunately they do not state which studies they are referring to.

Morgan and Russell (1975) found that poor pre-morbid family relationships predicted poor outcome 4 years later ($r = .52$). They did not find any association between outcome and history of mental illness in the family, anomalous family structure or sibling rivalry. The same sample of patients were followed up at 20 years using the same methods with much the same results (Ratnasuriya, 1991). Hsu et al (1979) followed up 100 female patients 4-8 years after initial presentation and found poor parental relationships predicted poor outcome. All these studies were retrospective casenote reviews with attendant problems of rater bias. However, Hall et al (1984) found no relationship between outcome and family relationships. In this study, unlike the previously mentioned ones, the initial assessments were independent from outcome assessments, thus avoiding possible rater bias.

In a study of 78 patients using the same procedure as Hsu (Morgan et al, 1983), the authors noted that the disturbance predictive of poor outcome was one of hostile attitude "strikingly similar to the relationships between high negative expressed emotion in families to poor outcome in schizophrenia". The role of Expressed Emotion in Anorexia Nervosa will be discussed in Chapter 4.

Crisp et al (1974) measured the mental health status of mothers and fathers at initial assessment and found a significant correlation with outcome.

In a study of younger AN patients, Bryant-Waugh et al (1988) found that children from one parent families or with a parent who had been married before had a poor prognosis. In a study of male AN patients Crisp et al (1986) found that poor parental relationship predicted poor outcome.

While initial poor family functioning appears to be a good indicator of prognosis, no inference can be drawn on the direction of causation. None of these studies controlled for the initial severity of illness which could be a confounding factor. Confounding factors are particularly problematic when the Morgan and Russell scales are used as the measure of illness severity as the scales include an assessment of family functioning.

2.13 The Influence of Bulimia on Family Functioning

Studies have found that there is a greater prevalence of impulsive behaviour such as stealing, alcohol and drug abuse, self mutilation and suicide attempts in AN patients who binge versus those who do not (Casper et al, 1980; Vandereycken and Pierloot, 1983; Eckert et al, 1987). Several studies have sought to investigate whether there is also a difference between the **families** of patients in whom a varying degree of bulimic behaviour is present compared to AN families. Thus families of AN restrictor patients have been compared with AN-bulimics and AN families have been compared with bulimia nervosa families.

Most self-report studies of family functioning find that the presence of bulimia is a sign of greater conflict in a household (Strober, 1981; Garner et al, 1985; Kog et al, 1985b; Kog et al, 1989; Stern et al, 1989; Shisslak et al, 1990). The exception is Waller and colleagues (1990). Most studies show that BN sufferers reported less cohesive families than AN sufferers

(Garner et al, 1985; Stern et al, 1989 v Kog et al, 1985; Kog et al, 1989; Waller et al, 1989, 1990; Shisslak et al, 1990) but only 1/4 found parents reporting less cohesion (Strober, 1981 v Kog et al, 1985; Stern et al, 1989; Waller et al, 1989, 1990).

Where expressiveness and adaptability could be measured, no differences were reported between parents (Strober, 1981; Waller et al, 1989, 1990; Stern et al, 1989). 2/3 studies found bulimic sufferers showing more difficulty with expressiveness (Garner et al, 1985; Stern et al, 1989 v Waller et al, 1990) and in 2/4 studies bulimia sufferers had more problems with adaptability (Garner et al, 1985 v Stern et al, 1989; Kog et al, 1989; Waller et al, 1990). Bulimic patients have reported more violence and disagreements between their parents, more substance abuse, criminal convictions, sex abuse and social instability (Piran et al, 1988).

Moving from self report studies to the interview study of childhood experience, Schmidt et al (1993) found that their bulimia nervosa patients were twice as likely to report family adversity as were their anorexic nervosa patients.

Prevalence studies using family study methodology have produced equivocal results on the restricter v binger question. Strober et al (1982) reported higher rates of alcoholism in the families of bulimic-anorexic patients compared to restricter families but this was not confirmed by a similar study (Halmi et al, 1991). Halmi et al (1991) also failed to confirm Strober et al's (1982) finding that depression was more common in the restrictor families. However, they did find that mothers of bingers had more anxiety disorder.

A cautionary note needs to be sounded here. Welch and Fairburn (1993) in a community survey found that only 15% of bulimia nervosa sufferers were known to treatment agencies. The major difference between known and unknown sufferers was that the known group had more personality

problems, family problems and other psychiatric problems. Sampling bias may thus explain many of the observed differences.

2.14 Summary

1. The cardinal feelings of Anorexia Nervosa are described with an emphasis on observed behaviour. (2.1.1 to 2.1.2)

2. AN patients have a high level of general psychopathology or neurotic symptoms and are reported to have a high rate of personality disorder but not in excess of other psychiatric disorders. In common with other neurotic patients, self esteem is low. (2.1.3 to 2.1.5)

3. They are reported to have difficulties in social functioning and psychosexual functioning but less difficulties in education and employment. They are also reported to have difficulty in separating from the parental home and gaining autonomy. However, these observations have not been tested against norms for their well peers. (2.1.6 to 2.1.9)

4. The natural history and epidemiology of the illness demonstrates it to be a widespread difficulty with a grave outlook for 25% of sufferers. (2.2)

5. The multiple aetiological theories about the illness and the numerous treatment packages that have been tried show that it is still a perplexing and difficult to manage problem. (2.3 to 2.4)

6. Attitudes to the families of AN patients have shifted over time. A dominant influence in recent years has been Minuchin's family systems theory and his concept of the "psychosomatic family". (2.5)

7. Current attitudes to families were explored by describing the practices of several major treatment centres in Europe and North America. (2.6)

8. The only demographic differences between AN families and well families that have stood up to the scrutiny of controlled research studies is the finding that incidence is higher in Social Class I and II than would be expected from chance. The class skew is not, however, as marked as it once was and was not found in the Swedish community study. (2.8)

9. The prevalence of eating disorders in the families of AN patients is 4 to 8 times higher than would be expected by chance. However, over 90% of families do not have a family member with a history of eating disorder. There is no support for the lesser criterion that anorexic families are particularly preoccupied with weight, shape and eating behaviour. (2.9.1)

10. Rates of psychiatric illness in anorexic families are 2 to 3 times higher than would be expected by chance. Most of the studies have focused on affective disorders. Rates are higher for mothers than fathers. (2.9.3)

11. While there is anecdotal evidence of personality difficulties in parents, in the only controlled study of personality disorder known to the author, the matched control mothers had more personality disorders than the anorexia nervosa mothers. (2.9.4)

12. There is no evidence to support Hilde Bruch's assertion that the childhoods of AN patients were rigidly controlled and overprotected by parents. There is more evidence for the opposite assertion: that high parental indifference and lack of control were present. (2.9.5)

13. Self report studies reveal that AN patients and their parents do not see themselves as being any more or less cohesive or any more or less adaptable than control families. The evidence on conflict and expressiveness remains equivocal with about half the studies showing raised levels in AN families. (2.10.1)

14. Controlled interview and observation studies have not found AN families to be more cohesive (overprotected or enmeshed) than well families. They have been found to be either as conflictual or less conflictual than well families. (2.10.2 to 2.10.3)

15. Family therapy has been found to be more effective than individual therapy in younger, less chronic patients provided that weight has already been restored after treatment. (2.11)

16. As with most psychiatric disorder, poor family functioning predicts poor long term outcome. (2.12)

17. Patients with bulimic symptoms have a more negative view of their families than those without, particularly in the arena of conflict. The **parents** of patients with bulimia have a less negative outlook than their daughters. (2.13)

CHAPTER 3: LITERATURE REVIEW - CYSTIC FIBROSIS

3.1 Aetiology and Clinical Features

Cystic fibrosis (CF) is a life long inherited disease characterised by abnormal secretions of the exocrine glands. The abnormal secretions result in a variety of secondary clinical effects including chronic lung disease and pancreatic insufficiency. Widespread recognition of the illness is recent and it was not until 1938 that a major detailed description of the disease was given (Andersen, 1938).

CF has an autosomal recessive pattern of inheritance, the heterozygote rate being about 1:25 in the United Kingdom. This means that when both parents are carriers, there is a 1 in 4 chance of the offspring of that union having the disease. In 1989 researchers at Toronto in Canada and the University of Michigan in the US isolated the genetic locus of cystic fibrosis (Rommens et al, 1989). The most common CF mutation accounts for about 80% of the 2 million CF carriers in Britain (Watson et al, 1992). The remaining 20% include a considerable variety of differing mutations. It is thought that the nature of these mutations determine the severity of the illness (Report of the Royal College of Physicians, 1990).

The nature of the basic defect is not yet fully understood but has been narrowed down to a defect in the control of chloride ion channel function at the luminal surface of epithelial cells lining the airways, the sweat ducts and pancreatic ducts. Most of the varied and widespread clinical manifestations of CF are due to the production of abnormal sticky secretions by the exocrine glands. In 90% of cases, thick secretions in the pancreas block normal delivery of enzymes leading to impairment of digestion and absorption and distinctive foul smelling stools if untreated. Abnormal secretions in the bowel can cause obstruction. At birth lungs are normal but are prone to infection which stimulates the secretion of profuse amounts of sticky mucus. There is a tendency for the mucus to stagnate

predisposing to further infection and secondary lung disease which eventually becomes irreversible. As the child gets older, infection becomes chronic and causes progressive damage to the lung structure. Eventually death results from a combination of respiratory and cardiac failure.

To the casual observer CF patients look quite normal. A persistent cough may be all that is noticed. However, because of malnutrition, short stature, low weight and delayed puberty are more common than in the general population. Where lung disease is severe, patients tend to be barrel-chested but this is well camouflaged by normal clothing. Intellectual functioning is normal and children should be educated in ordinary schools.

3.2 Prognosis

There is a great variability in both the mode of presentation and the severity and progression of the illness. 80% of sufferers are diagnosed within the first 2 years of life. A few people with very mild or atypical variants of the illness may reach adulthood before diagnosis.

In 1948 no known CF patient survived into adolescence but by 1966 the median age of survival was 12 years. Today median expectations of life are usually cited as 20 to 25 years. These data refer to patients who were first treated 20 or more years ago. The child who is diagnosed today should expect to survive well into adulthood without severe lung disease and with a greatly improved quality of life (Levison et al, 1987; Britton and Knox, 1991). The estimate for 1990 was that there are at least 2000 CF patients over the age of 15 in the UK (Report of the Royal College of Physicians, 1990). Some of the improvement in survival curves may be due to improved diagnostic efficiency leading to milder variants of the disease being recognised and thus boosting the survival statistics. However, several factors are thought to contribute to the improved outlook: the establishment of specialist treatment centres, the use of antibiotics, improved nutrition, more effective physiotherapy regimes and possibly

earlier diagnosis and commencement of treatment (Goodchild and Dodge, 1989). If available treatment is used consistently and optimally, many patients can achieve a near normal lifestyle for many years with normal school attendance and social life.

3.3 Treatment

From the time of diagnosis, CF is an illness that has to be carefully managed with a major investment of vigilance and care by the Health Service. Because the treatment regime is largely home-based, it also requires a major investment of discipline and compliance from both the patient and the family. The resourcefulness, independence, expertise and responsibility that families and patients develop in the course of achieving this means that the doctor/patient relationship becomes far more one of equal partnership than is customary for most forms of healthcare. Not infrequently they know more about the illness than do their medical and other professional advisers (Report of the Royal College of Physicians, 1990).

3.3.1 Nutritional Problems

Malnutrition and failure to thrive are closely associated with accelerated decline in lung function which in turn adversely affects overall survival (Durie and Pencharz, 1989). In order to maintain growth and nutrition, the majority of CF patients need to take pancreatic enzyme preparations with every meal and daily doses of vitamins. Parents have to learn to adjust the dosage according to the composition of stools rather as a diabetic learns to adjust insulin intake to food consumption. Once patients reach school age they are expected to manage this themselves. In order to maintain normal growth and body weight, CF patients may need to increase calorific intake to 120% to 150% of normal with a high calorie, high protein diet so patients are under pressure from family and doctors to keep eating. A few patients also need nutritional supplements or special formulae foods.

When patients are well, the dietary requirements are not usually a problem but, as loss of appetite is a common symptom of fever, periods of severe infection are potential battlegrounds for eating problems. In addition daily extra vitamins have to be taken.

3.3.2 Respiratory Problems

> "Adequate and consistent physiotherapy is possibly the single most important factor in preventing chronic lung infection and in aiding antibiotics to eradicate infection."
>
> Goodchild and Dodge, 1989

Physiotherapy needs to be carried out for 20 minutes or more periods once, twice or three or more times a day according to need. It is used as a prophylactic measure as well as a treatment procedure to ensure adequate drainage of the lungs. In the early years, the duty to carry this out is discharged to the patient's parents who are trained in the appropriate techniques by a physiotherapist. As the child approaches adolescence, he is taught how to clear the lungs himself and parents are encouraged to hand over this responsibility to the child. How far parents are successful in this is often seen as a yardstick of whether parents, particularly mothers, are overinvolved with the child. Compliance with the physiotherapy regime often goes down in the teenage and adult years (Czajkowski and Koocher, 1986). A recent study found less than half CF adults believed physiotherapy to be effective and skipped at least one quarter of their physiotherapy (Fong et al, 1990). If a vigorous fitness regime is followed, physiotherapy can occasionally be dispensed with.

Antibiotics to combat lung infection have contributed greatly to longer survival and improved quality of life. However, once established, infection is very difficult to control and some pathogens are impossible to eradicate. There is no universal agreement by doctors on how and when antibiotics should be administered. Some centres give continuous antibiotic treatment but others only as a response to raised infection. CF patients and their

parents become expert in recognising the signs of infection, in recognising when they are "ill" and when they are "well" and when action needs to be taken by reporting to their physician for sputum tests and possible treatment. Many of the more potent antibiotics have to be administered intravenously. Traditionally this has involved admission to hospital but an implantable reservoir (Porthacath) has enabled this to be carried out at home by the families themselves. This procedure requires much attention to detail and discipline. In the more advanced stage of the illness, supplementary oxygen to assist breathing may be needed. Again cylinders can be delivered to the patient's home to avoid hospital admission.

3.3.3 Hospital Admissions

Patients are expected to attend out-patient clinics for routine regular check ups which could be set at anything from 1 to 6 months depending on age and health status. Hospital admissions are kept to a minimum and many CF patients have never been in hospital at all. There can be miscellaneous complications such as intestinal obstruction, arthritis, diabetes and liver failure which also have to be treated. In advanced stages of the illness major haemoptysis (coughing up blood) and complications of advanced lung deterioration such as pneumothorax (collapsed lungs) will all require admission to hospital.

Until recently CF had to be regarded as an ultimately fatal illness. However, in 1984 the first heart-lung transplant was carried out. The patient died at 8 weeks. For a few patients the operation appears (so far) to be a dramatic success but the number of donor lungs is unlikely ever to satisfy demand, the operation remains high risk and the lottery of waiting and hoping is highly stressful to family and patients alike. Warner, commenting on outcome studies at the Great Ormond Street and Brompton Hospitals states that only a sixth of the referrals had truly benefitted from heart-lung transplantation and concludes

> "The psychological problems which occur in the families drawn into the transplantation programme are immense. For some the hope generated has a very positive effect but for many the whole procedure is a nightmare that ends in disaster"
>
> Whitehead et al, 1991b

3.3.4 Future Prospects

On the horizon is the possibility of gene therapy in which a normal gene could be introduced, possibly via aerosol sprays into the genetic material of the lungs to overcome the effects of the CF mutation (Report of the Royal College of Physicians, 1990). The recent success in developing a mouse model of CF by targeted insertional mutagenesis should spur on progress in this area (Dorin et al, 1992).

Localisation of the defective CF gene has led to the development of a simple mouthwash screening test that can identify the most common mutations. This has opened up the possibility that in the future fewer children with CF could be allowed to be born but whether a screening programme for carriers will be introduced nationally will depend on political, financial and ethical consideration (Britten and Knox, 1991; Editorial, The Lancet, July 1992).

3.4 Psychosocial Functioning of the CF Patient

3.4.1 Childhood Studies

I have arbitrarily defined a "childhood" study as one in which the mean age of the sample is 12 or less. This categorises reports such as Steinhausen et al's 1983 study (mean age 10, range 5-18) as a "childhood" study which the reader may find troublesome.

Because until recently life expectancy was so limited, most of the earlier studies on cystic fibrosis focused on childhood. Single group studies without comparisons were carried out by Lawler et al (1966), Spock and Stedman

(1966), Kulczycki et al (1969), McCollum and Gibson (1970), Tropauer et al (1970), Cytryn et al (1973), Allan et al (1974), Burton (1975), Falkman (1977), Drotar (1978). While data collection procedures varied, each study included an interview with a child, parent or both. In some cases projective tests were included. The descriptions of the CF patients in many of these studies are very rich and detailed. They present a rather dismal picture of behaviour problems, anxiety and lack of independence and date from a period when the quality of life for CF children was considerably worse than it is today. However, the limitations of the methodology suggest that they should be accepted with reservations.

Where CF children have been compared to a control group or with an assessment procedure for which norms are available, most studies have found that the children are free of serious psychological damage and indistinguishable from well children (Cull, 1974; Tavormina et al, 1976; Gayton et al, 1977; Drotar et al, 1981; Cowen et al, 1985; Simmons et al, 1987; Kashani et al, 1988; Cappelli et al, 1988, 1989a). Pumariega et al (1990) reported higher levels of disturbance in children with neurological problems than with cystic fibrosis. These studies used self-report measures, usually completed by mothers and occasionally fathers or teachers. Some studies also used interviews.

However, 2 controlled studies have been carried out using standardised psychiatric interviewing techniques which have come to different conclusions from the self report work. In a study in Germany (Steinhausen et al, 1983) a sample of CF children and control groups of asthmatic and healthy subjects were given structured interviews. 53% of the CF children were given ICD psychiatric diagnoses compared to 28% of the asthmatic children and 17% of the well children. Emotional disorders, particularly separation anxiety, were the most common CF diagnoses. There was no difference between groups for conduct disorders. This finding was confirmed by checklists completed by school teachers and parents. Thompson et al (1990) using the DSM-III classification gave 58% of their

American CF patients a psychiatric diagnosis compared to 77% of psychiatrically referred children and 23% of normal schoolchildren. Again anxiety was the most common diagnosis. Prevalence of conduct disorder was low. Unfortunately neither study describes the physical health status of the CF sample.

Also using interview method but with the much briefer Psychiatric Screening Inventory, Breslau and Marshall (1985) compared a good sized sample of CF children (n = 66) with children with cerebral palsy, myelodysplasia, multiple physical handicaps and well controls and found no difference in rate of severe psychiatric disorder for CF children compared to well controls. However, they did find more mild anxiety and behaviour problems in CF children. They followed their sample up 5 years later and found that the CF patients had improved significantly.

A recent single group study on child patients being assessed for heart-lung transplantation found only 45% of this very ill group of patients with normal psychological functioning when rated for psychological functioning by a psychiatrist and a psychologist (Whitehead et al, 1991a). This high rate of disorder may of course say more about the stresses of being included in a heart-lung transplantation programme than it does about the patients (see Section 3.3.3).

3.4.2 Adolescence and Adulthood

One of the earliest descriptive accounts of adolescents was that of Teicher (1969) who reported that these patients were suicidal, depressed, angry, hopeless and isolated. Many had renunciated all treatment and declared a wish to die quickly. Boyle et al (1976), using psychiatric interview and projective tests, reported "much anxiety and depression" in a group of adult and adolescent patients. They attributed stress to problems concerning physical appearance, problems in parental relationships, particularly

infantalisation by mothers, and increased concern about the future. Neither of these studies had a control group.

Several investigations have been carried out using self report questionnaire methodology either with a comparison group or with available test norms. Kellerman and colleagues (1980) found no difference in anxiety levels between CF adolescents and large comparison groups of other physically ill groups and normal healthy controls. A large scale study from Toronto (Simmons et al, 1985) of CF adolescents found no difference in the rate of behavioural disturbance (as measured by a parental rated check list) compared to rates in the Isle of Wight normal population survey (Rutter et al, 1976). Cappelli et al (1989a) found no difference in levels of depression between CF, diabetic and normal adolescents. Steiner et al (1990) compared 10 CF adolescents with 10 Anorexia Nervosa patients matched for age and weight and found less trait anxiety in the CF sample.

The Toronto group also looked at their population of CF patients over 16 (Cowen et al, 1984). Patients completed the General Medical Index. The severity of emotional disturbance scores approached those of medical patient populations (ie raised) rather than normal populations. Pearson et al (1991) found higher rates of depression and anxiety in a group of CF patients aged 16 to 40 than she would have expected from the general population. However, it should be noted that response rate of her sample was only 48%. Results from these self report questionnaire studies suggest that CF adolescents do not differ from normal adolescents in levels of psychopathology but by adulthood some divergence has taken place.

Turning to interview studies with some control data, Bywater (1981) interviewed 27 British CF teenagers and found that 63% of the patients admitted to "feelings of misery" during the previous year. This compared with a rate of 45% in normal teenagers in a large epidemiological study who were asked the same question (Rutter et al, 1976). She did not assess formal psychiatric illness.

A "structured clinical interview" found no difference in prevalence of depression in a small study of CF adolescents and matched normal control adolescents (Straker and Kuttner, 1980). Moise and colleagues (1987) interviewed CF adults with the Psychiatric Screening Inventory and found no increase in disturbance compared to test norms. To date no studies have been carried out on CF adults and adolescents using ICD or DSM interview based criteria of illness.

3.4.3 Cystic Fibrosis and Eating Disorders

It has long been known that chronic undernutrition in CF is correlated with accelerated decline in pulmonary function which in turn adversely affects overall survival. Malnutrition occurs from energy imbalance which results from 3 inter-related factors: increased energy losses, increased energy expenditure and reduced energy intake. Faecal nutrient losses from maldigestion/malabsorption can be compensated for to a large extent by ingesting pancreatic enzymes. However, many patients continue to have severe steatorrhoea and azotorrhoea. Energy expenditure increases due to the increased work of breathing in patients with advanced lung disease but increased energy expenditure (compared to well subjects) has also been observed in patients with good lung function for reasons which are not understood.

To compensate for energy losses it is widely accepted that energy intake should usually exceed normal requirements and crude estimates have suggested that patients require 120-150% of the recommended daily allowance. However, adolescents with CF have frequently been found not to reach these targets Bell et al, 1981). There are a number of complications that limit appetite and oral intake in advanced disease. Oesophagitis is frequently associated with pain, anorexia and vomiting following bouts of coughing. The distal intestinal obstruction syndrome causes recurrent abdominal pain and patients often find that the symptoms are exacerbated by eating. Severe chest infections induce anorexia and

often give rise to vomiting which may further reduce intake (Durie and Pencharz, 1989; Goodchild and Dodge, 1989).

Psychological factors in the reduced energy intake of adolescents should also be considered. Durie and Pencharz (1989) comment on the need to be alert to the presence of clinical depression as a cause of loss of appetite. An anecdotal account (Pumareiga et al, 1986) described 13 CF adolescents with eating difficulties (from a total clinic population of 108 CF adolescents). None of these patients met DSM-III criteria for Anorexia Nervosa although the authors reported that DSM-III "atypical eating disorder" could describe this group. Unfortunately information about the sample is sketchy and crucial information about whether patients were engaged in the "relentless pursuit of thinness" or were over pre-occupied with weight and shape is absent. The authors comment that most of the families demonstrated Minuchin's psychosomatic family characteristics but give no details of how this was measured. There could be alternative explanations for weight loss. Half the sample met diagnostic criteria for major depressive disorder which could explain loss of appetite. Lung function of the group was noticeably low (mean vital capacity 38%) which suggests that many of the complications of advanced disease described above could be at work.

Pearson et al (1991) gave a self report checklist (Eating Attitudes Test) to CF patients. They found that 16% of CF children (mean age 11) reached the cut-off for anorexia nervosa and 3% of young adults (mean age 25). It is unfortunate that they did not go on to interview the patients as answers to many of the questions on the EAT (eg resistance to eating and pre-occupation with food) could have a non-psychological interpretation in a CF population. Steiner et al (1990) compared a small group of AN patients with CF patients matched for weight. On the Eating Disorder Inventory, CF scores were indistinguishable from the norms of well subjects and significantly healthier than the AN subjects. Parents of the subjects were also asked to rate their children on the Slade Anorexia Behaviour Scale. AN parents were at the 99th percentile and CF parents at the 58th

percentile. Items most likely to be endorsed by CF parents were those concerned with resistance to eating such as "picks at food" and "begins by cutting food into small pieces".

As the evidence stands, it seems likely that most eating difficulties in CF adolescents are determined by their physical health status. However, co-morbidity should not be ignored and depressive illness needs to be considered. Furthermore, normal prevalence of eating disorders should not blind the physician to their existence. Goldbloom (1988) describes a single case of a CF patient with the bulimia subtype of anorexia nervosa. It was depression, not AN, that led to the initial psychiatric referral.

3.4.4 Denial of Illness

The finding that when self report methods are used, CF patients tend to be indistinguishable from their peers may reflect the true state of affairs. Alternatively, parents (who by and large reported on children) and adolescents and adults (who reported on themselves) could be denying illness. Suls and Fletcher (1985) have observed that a patient threatened by major illness may show two defence reactions. The first is denial, an avoiding response which focuses attention away from the illness. Patients using this mechanism show little direct interest in their illness and symptoms. They may assert the need "not to give in" to the illness. The alternative defence reaction is to respond with increased vigilance, the sensitising response. These patients closely monitor their physical status, may be concerned with future disability and openly talk about their plans and will be aware of recent research progress in the illness. Bronheim (1978) states that "in order to function, the patient must deny death but not the disease". In his review of adult CF patients, Aspin (1991) suggests that CF patients may fare better when using avoidance rather than attention responses, citing the work of Strauss and Wellisch, (1981); Cowen et al, (1984); Moise et al, (1987) to support his case. Denial seems to be unrelated to health status.

3.4.5 Age and Individual Psychopathology

Do psychological problems increase with age? Pearson (1991) compared a sample of 8-15 year olds with a sample of 16-40 year olds and found rates of depression and "global disturbance" to be higher in the older group. Cowen et al (1984) found less disturbance in patients aged 16-19 compared to those over 20 and interpreted this as a reaction to imminent profound physical disability and death. Strauss and Wellisch (1981) report that patients more than 23 were more likely to be uncomfortable in social situations, to have self doubts and to be preoccupied with physical function. However, rates of psychiatric disorder also increase with age in the normal population (Rutter et al, 1976).

If there is an increase of psychopathology with age, this cannot be explained by deterioration in physical health status as most researchers find no link between severity of illness and mental health or behavioural problems. This seems to be true for children (Cytryn et al, 1973; Drotar et al, 1981; Breslau, 1985; Brennan et al, 1990; Pearson et al, 1991; Whitehead, 1991a; Sanders et al, 1991) and adolescents and adults (Cowen et al, 1984; Simmons et al, 1985; and Sinnema et al, 1988). The one exception to this finding was the German interview study of Steinhausen et al (1981, 1983) where a correlation was found between lung function and psychiatric illness although not for two other indicators of physical health.

3.5 Self Esteem

Low self esteem is so commonly found in psychiatric patients that it has been used as an indicator of psychological well-being. Self esteem is customarily measured by self report questionnaires. Kellerman et al (1980), Cowen et al (1984), Simmons et al (1985), Moise et al (1987) and Shepherd et al (1990) found no difference in self esteem between their CF adolescent or adult samples and either normal controls or normative data.

Smith et al (1983) found that his CF sample scored at the 30th percentile of normal. Self esteem seems robust in CF patients.

3.6 Other Indicators of Psychological Health

How well do CF youngsters adapt to the tasks of adolescence and adulthood? Mearns (1986) has described some of the problems that can face CF teenagers in terms of the developmental tasks which should be completed during adolescence.

3.6.1 Independent Self Care

Mearns describes the first of these tasks as making the change from being cared for to being able to perform the tasks of daily living and to learn to care for others. Maintaining good lung function is vital to the well being of the CF patient and the severity of the disorder at adolescence will dictate the degree of physical independence the patient can achieve. Since the introduction of forced expiratory breathing and coughing as a method of chest drainage, older patients have been able to do physiotherapy independently of a helper and are thus free to develop a lifestyle separate from their parents. A study in Holland (Sinnema et al, 1988) that merits particular attention because of the large sample size (n = 64 CF patients aged 12-20) and the inclusion of several matched control groups looked at various measures of independence. They found CF patients less independent on some measures of personal care, eg when to put on clean clothes, when to wash hair, but not others, eg decisions about bedtime and being warmly dressed.

3.6.2 Social Functioning

Another task of adolescence is to form lasting relationships, particularly peer group relationships. Shepherd et al (1990) found no difference in the social network density of their sample of CF adults and a healthy

comparison group, measuring such variables as frequency of visits with friends, number of close friends and number of voluntary groups, clubs and organisations belonged to. A similar picture was found in Bywater's uncontrolled British study (1981). By contrast, Lindstrom and Kohler (1991) working in Nordic states found reduced social networks for CF and myelomeningocele teenagers. Sinnema et al (1988) found CF teenagers were less active outside the home but found no dissatisfaction with this situation and no reported wish to change.

3.6.3 Psychosexual Functioning

In assessing psychosexual functioning, one or two complicating factors need to be mentioned. The likelihood of sterility in the male is very high and tests to determine this may not be possible until puberty is complete. For the female, although pregnancy may be possible, it is not always advisable and even in the fittest may lead to deterioration in lung function (Mearns, 1986).

Pubertal delay and short stature are more common in the CF population than in the general population. This appears to be a feature of malnutrition rather than intrinsic to the disease (Landon et al, 1984; 1987). The Dutch study (Sinnema et al, 1988) of CF adolescents found a delay in the development of intimacy and sexuality compared to a sample of asthmatic and well subjects. It also found that the mediating variable was pubertal delay. Landon and Rosenfeld (1984) compared CF males with 2 healthy control groups, one with short stature and delayed puberty and one without. Self esteem was linked to short stature and delay, regardless of whether the subjects had CF or were healthy.

Cromer et al (1990) explored the knowledge, attitudes and behaviour related to sexuality in a group (mean age 17) of CF patients, myelomeningocele patients and matched controls and found 28% of the

myelodysplastic group, 43% of the CF group and 60% of the well group reported previous sexual activity.

There appear to be differences in psychosexual adaptation between boys and girls. Looking at a sample of single CF patients aged 19 or more, Coffman et al (1984) found that female CF patients were assessed by the researchers as having problems of sexual adaptation compared to well subjects using both a conservative and a liberal definition of sexual adaptation. They began dating later, dated less often, felt less attractive, had less sexual desire and had more sexual problems than physically healthy females. The single male patient with CF seemed to fare better than the females and approximated the healthy male group. The same research group had earlier reported on the sexual adaptation of a group of married CF patients and found that most had good or excellent sexual adaptation (Levine et al, 1982). Zeltzer et al (1980) found in their large sample of adolescents with several physical illnesses including CF that females in all groups reported more impact of illness on attitudes to physical appearance than did males.

It is likely that severity of illness is a powerful mediating factor in sexual adaptation. In Coffman's study (see above) the male sample had markedly better lung function than the female sample. The married CF patients reported on by Levine (see above) also had better lung function than the unmarried groups. Sinnema's study (see above) found severity of illness was related to sexual delay in girls but not in boys.

In spite of the difficulties, 82% of the CF females in Coffman's study (1984) described themselves as being satisfied with their sexual lives. Shepherd et al (1990) found no difference in levels of satisfaction for sexual activities between CF adults and healthy peers.

3.6.4 Education and Work

A further task of adolescence is to develop a commitment to work and to acquire skills to become materially self supporting. CF children are of normal intelligence and should be educated in normal schools. It has frequently been commented on that the educational achievements and work records of CF patients, sometimes despite severe disease, is remarkable (Bywater, 1981; Strauss and Wellisch, 1981; Mearns et al, 1986; Penketh et al, 1987; Shepherd, 1991). Di Sant'Agnese and Davis (1979) described 85% of their adult patients as being involved with school, career or housework. Cowen et al (1984) reported that 93% of their male and 89% of their female patients over 16 were in school or higher education. A Dutch study (Sinnema et al, 1983) found stay on rates at school much higher in their CF population than in the general population. However, unemployment levels were higher in the CF population than in the general population particularly in manual occupations. In Britain, inflexibilities in the Social Security system mean that patients are not able to work part-time without losing sickness benefits. Visits to clinics and time off through illness are much easier to accommodate at school and college than in paid employment. Goldberg et al (1985) looked at school and occupational status of patients 6 years after the assessment of IQ, self esteem and clinical health. They found that severity of illness was the best predictor of school status. There is thus no evidence that anything other than physical health is keeping CF patients out of the job and education market.

3.6.5 Autonomy and Leaving Home

Given that there are bound to be marked cultural differences in how and when young people leave home (Goldscheider and Waite, 1987), this is a rite of passage for young adults. Strauss and Wellisch (1981) found that two-thirds of their adult CF sample no longer lived with parents, the most common reason for staying being financial or being too sick to live alone. Reporting on a large series at the Brompton Hospital, Penketh et al (1987)

found that most patients remained single usually living with parents. In Toronto Cowen et al (1984) report that 64% of their male patients over 20 and 83% of their female patients were married or no longer living with their parents. None of these authors supply population norms. Sinnema et al (1983) found rates of adolescent and young adult CF patients living in the parental home to be the same as for the total population of the Netherlands.

3.7 Coping Skills of the Patient

Another indicator of psychological function is "coping", the measure of a family's ability to meet the demands of life stresses. This is a complex mixture of personality structure, family influence, life events and factors related to the disease itself (Pinkerton et al, 1985). Coping has been extensively studied in CF **children** where complex interactions between family functioning, severity of illness and coping styles and compliance with treatment have been found (Venters, 1981; Patterson, 1985; Mollering, 1986; Cappelli et al, 1988; Patterson et al, 1990; Koocher et al, 1990; Sanders et al, 1991). The focus of all these studies is on the parents who are generally responsible for implementing medical procedures rather than the young CF child himself.

In a British study Pinkerton et al (1985), adult patients were split into copers (showing good adjustment to CF), and non-copers (poor adjustment) and intermediate copers by 5 independent clinical raters. They found that poor domestic environment (family relationships), malfunction in sexual relationships and emotional distress (anxiety, depression, worry etc) effectively differentiated between the 2 groups. The non-copers had more admissions to hospital and lower compliance. However, they found, as have others, (eg Sanders et al, 1991) that coping had no relationship to lung function status.

3.8 Are There Demographic Differences between CF Families and Other Families?

3.8.1 Social Class

The genetic inheritance pathway of CF would suggest that there should be no social class bias in birthrates. However, a recent descriptive account of the characteristics of patients attending the Brompton Hospital, London, a major specialist centre, noted that 56% of their adolescent and adult patients came from social classes I and II against an expected proportion of 20% (Penketh et al, 1987). Survival rates are known to be higher in patients managed in specialist centres (Goodchild and Dodge, 1989). The extent of the effect of social class on survival in cystic fibrosis was investigated by Britton (1989). He found a strong independent effect for social class on age at death. This is a fairly common finding in chronic disease. The middle classes seem more effective at seeking out and maximising the best clinical care. They have better resources to permit visits to distant hospitals, to buy better quality equipment and to supplement diet. There may also be factors such as increased parental smoking, poor quality or overcrowded damp housing and lower levels of education in CF households that lead to early death in lower class CF patients (Black et al, 1982).

3.9 Is There More Individual Psychopathology in CF Family Members?

3.9.1 Mothers

In early childhood in CF as in all chronic illness, the heaviest burden of care usually falls on the mother. It is her freedom that is curtailed by the illness, she who takes the child to the GP and clinic, who takes on the main responsibility for the daily care routine (Cull, 1974; Burton, 1985; Dolan, 1989; Nelson, 1989). Various studies have suggested that the mothers of

CF children suffer from depression, somatic complaints, and feelings of guilt and inadequacy (Turk, 1964; Lawler et al, 1966; Meyerowitz and Kaplan, 1967; Kulczycki et al, 1969; Tropauer et al, 1970; McCollum and Gibson, 1970; Allan et al, 1974). However, none of these early studies had a control group or use assessment methods with normative data.

Formal psychiatric interviews with control groups and ICD or DSM classification of illness are lacking. In Germany, Steinhausen et al (1981, 1983) carried out structured interviews with mothers of CF, asthmatic and well children. In their 1981 paper, they report that CF mothers suffer more depression and anxiety than the well control group. In the 1983 paper, reporting on the same sample, the differences between groups did not reach significance. These conflicting results may be explained by the use of the chi-square statistic in the 1981 paper and the F ratio in the 1983 paper.

Turning from psychiatric "caseness" to the less stringent criterion of "emotional disturbance", there have been several controlled studies using self report questionnaire methodology. Cull (1974) found more anxiety in a sample of mothers of CF patients compared to mothers of coeliac disease patients. 79% of her mothers also reported depression for which half had sought treatment. Gayton et al (1977) found raised incidence of depression. A more recent study, Walker et al (1987), found no difference in levels of depression between a group of CF mothers and mothers of healthy child comparison groups nor did they find any difference in reported levels of stress between the 2 groups. The Schwachman scores (a commonly used index of CF illness severity) of Walker's group were relatively high suggesting a fairly healthy CF group which may partly explain this difference. Nagy and Ungerer (1990) found CF mothers' mental health to be poorer than the general population.

What effects follow from mothers' mental health status? Where severity of illness has been measured objectively by clinicians or by lung function parameters, it appears not to correlate with mothers' psychological health

(Frydman, 1980; Holroyd and Guthrie, 1986; Walker et al, 1987). However, **mothers'** judgment of the severity of illness is correlated with her mental health and her perception of stress (Frydman, 1980; Breslau et al, 1982; Holroyd and Guthrie, 1986; Walker et al, 1987; Nagy and Ungerer, 1990). It will be recalled that most studies of the mental health status of young children used methodology that depended on reports of behaviour from mothers. The mental health status of the mothers could be a powerful mediating factor in the findings of emotional disturbance of those children.

While Cappelli et al (1989b) did not find differences in levels of overprotection in the mothers of CF children compared to well controls, they did find correlations between maternal overprotection and poor psychological functioning in CF children. This confirmed the similar findings using different methodology of Lewis and Khaw (1982).

The most stressful times for the parents of CF children are thought to be around the time of diagnosis and the stage of terminal illness (McCollum and Gibson, 1970). It is at least plausible that mothers would show more psychological disturbance at these 2 stages. The higher levels of maternal stress and psychological disturbance reported in the earlier studies probably reflect the fact that even in 1969 mean age of death was 12 years. More recent studies probably include more mothers in the long term adaptation phase of the illness.

All the studies cited so far have been concerned with pre-adolescent children. What is known about the mothers of adolescents and adult CF patients? By adolescence, many patients will be functioning more independently, will have taken over the management of physiotherapy and dietary supplements themselves and will be moving away from home. For others the terminal stages of their illness will necessitate dependency for nursing care and emotional support from their parents or, for those who have left home, their partners.

In an anecdotal account of 27 CF patients aged 13-30, Boyle et al (1976) stated that the patients were often overprotected and infantalised by their mothers. Bywater (1981) found that mothers of CF teenagers suffered more depression and general psychological malaise than was found in a study of normal 14 year olds' mothers using the same methodology. Her proportion of depressed mothers was the same as in a study of the mothers of children with spina bifida (Dorner, 1976). She found a link between severity of illness and maternal depression. No controlled interview studies have been carried out.

3.9.2 Fathers

Nelson (1989) has pointed out that fathers are shadowy figures in CF research. Because fathers tend not to attend CF clinics, few researchers have made the extra effort required to include them in research projects. There is abundant evidence that fathers take a much less active involvement in treatment than mothers (Cull, 1974; Burton, 1975; McCollum and Gibson, 1970; Falkman, 1977).

Cull (1974) found fathers to be no less anxious than would be expected from normative data. Depression does not seem to be a significant problem (Gayton et al, 1977; Bywater, 1981). Nagy and Ungerer (1990) found that fathers of CF children did not have mental health scores any different from the general population. They also found that support to wives by fathers of CF children in practical child rearing matters served to protect mothers from stress. Granvold et al (1990) found that where both parents were involved in treatment the CF child showed less anxiety than if only one (usually the mother) was involved.

3.9.3 Siblings

The impact of chronic disease on well siblings was studied by Breslau et al (1981). This was a large scale study in which several different chronic

illnesses were studied including CF. She found that the level of disability of the patient had no relationship to the adjustment of the normal sibling and found no difference in diagnostic categories for illness. Anecdotal accounts of siblings of CF patients have tended to stress not only the lack of negative impact on the well child but positive aspects of the sibling relationship as well: concern and helpfulness and protective care (Burton, 1975; Falkman, 1977; Mikkelson et al, 1978). Kerner et al (1979) found that following a death undue grief was unusual. In a study using standardised tests, Gayton et al (1977) found siblings to be well within the normal range of psychological functioning. By contrast, in the Toronto series (Cowen et al, 1986), while still within the normal range, CF siblings were slightly less healthy than their normal peers. Fanos and Nickerson (1991) interviewed adults whose sibling had died of CF. Age of the surviving sibling at the time of loss was the most significant determinant of later psychological problems, 13-17 being the most vulnerable age. The problems were guilt, anxiety, excessive concern for others and fear of intimacy.

3.9.4 Marital Discord

Begleiter et al (1976) reviewed the data of 14 American researchers and concluded that the incidence of divorce and separation was no higher among parents of chronically ill children including CF than among the general population. This was confirmed in a later review (Sabbath and Leventhal, 1984). In Sweden, Falkman (1977) found divorce was at a lower rate for her random sample of CF families than for the population at large. Walker et al (1987) found that 81% of their families with a CF member had both parents present in the home, whereas control families had 69% of both parents at home together.

There is evidence that CF can cause discord within a marriage but also for some families can serve to improve marital bonding (Sabbath and Leventhal, 1984). Both Cull (1974) and Allan et al (1974) reported that

some parents blamed CF for marital discord but others felt that the marriage had gained strength from mutual concerns. Burton (1975) found 57% of marriages "under strain" but, with no well control group, it is not clear how many ordinary marriages could be so described. Steinhausen and Schindler (1981) in a German interview study found more disagreement between CF parents on child rearing matters and fewer parental discussions than did well control families.

3.10 The Families of CF Patients

Research on families has tended to follow 3 trends: attempts to measure the impact or burden of the illness on the family, attempts to establish whether families have more "family pathology" or dysfunction than families of well children or children with other disabilities, and finally attempts to describe and evaluate the coping skills of those families.

3.10.1 Impact on the Family - Change Over Time

The impact of the illness on the family varies in quality and intensity over time. McCollum and Gibson (1970) have described 4 stages of adaptation to the illness: first the pre-diagnostic stage typified by anxiety and uncertainty. Venters (1981) found that the CF child's diagnosis was missed on 62% of the families first medical contact. Second is the acute crisis of diagnosis stage with antipathy, mourning, disbelief, anxiety and anger. Venters (1981) found that by the end of the first year after diagnosis, 71% of his families no longer felt that negative emotions dominated the day. The third stage is one of long term adaptation when families adapt to the routine of hospital appointments, physiotherapy, dietary management. The fourth stage is one of a grief reaction starting in the pre-terminal illness of the patient. Phillips et al (1985) found that the impact of hospitalisation was the most prevalent major problem for parents. Unfortunately, researchers only rarely state at which stage of adaptation the families are

at the time of the research project. Commonsense would suggest that responses to questionnaires and interviews could vary accordingly.

3.10.2 Socio-economic Impact

A survey in Norway where most married women are in employment found striking differences in the professional activity between CF mothers and matched control mothers. Most CF mothers had given up their careers. There was no difference in levels of professional activities for fathers (Michalsen et al, 1988). However, in America, Phillips et al (1985) found that 78% of parents interviewed did not feel that their career potential had been adversely affected by their CF child's condition. Meyerowitz and Kaplan (1967) reported that 54% of mothers were employed prior to diagnosis but only 26% after diagnosis. Hospital and clinic treatment of CF is now free of charge in many major treatment centres. Where it is not fully subsidised, the extra financial commitments are alarming. In 1971 McCollum estimated that in her American sample one-quarter of total family income was being spend on the illness. Even where treatment is free or subsidised, there are still financial burdens of transport, loss of earnings, special diets and better quality equipment (Turk, 1964; Kaplan, 1967; Kulczycki et al, 1969; Allan et al, 1974; Burton, 1975; Gayton et al, 1977).

The Questionnaire on Resources and Stress is a self report scale that attempts to measure the impact of childhood illness on family life. Walker et al (1987) found to her surprise no difference in reports of levels of burden between the mothers of well and CF children. She found that mothers' subjective severity of illness rates had a much closer relationship with assessment of burden than did objective severity of illness. Holroyd and Guthrie (1986), using the same scale, found considerably less burden reported than they had expected. Parents of children with neuromuscular disease reported much more burden.

3.10.3 Family Functioning

There have been several studies using self report measures of family functioning. Cowen et al (1986) reported on their large sample of Canadian CF patients (n = 298). They found that on 2 measures of family functioning, the Family Assessment Measure (Skinner et al, 1983) and the Family Adaptability and Cohesion Evaluation Scales (Olson et al, 1985), families had scores within the normal range. They reported an increase in family dysfunction with patients' age. Test norms are not available that could show whether this is a pattern in well families too. Lewis and Khaw (1982) also using the FACES found no difference between normal, CF and asthmatic families.

Shepherd et al (1990) assessed satisfaction with family functioning using a self report questionnaire and found no difference between a sample of CF adults and well controls.

Using the Family Environmental Scales (Moos and Moos, 1980), Patterson et al (1990) found that family functioning had no bearing on change in health status over a 15 month follow up period. Again using the FES, Johnson et al (1985) found that families where the CF child was later born were slightly more functional than families in which the child was first born. They found that the CF families showed more cohesion and less conflict than expected from norms of families with healthy children.

Turning to interview studies, the earlier anecdotal accounts reported family unit breakdown of communication, social isolation, decreased family satisfaction and poor adjustment (Turk, 1964; Meyerowitz and Kaplan, 1967; Kulczycki et al, 1969; Gayton et al, 1977). More recently, Bywater (1981) reported most of the CF teenagers she interviewed had good relationships with parents and that the family tended to do things together.

Kucia et al (1979) observed the families of well adjusted and maladjusted CF children carrying out a problem solving task and found that families of well adjusted children were able to suggest more ways of problem solving. The task (to discover the rules of a game) was not particularly salient to CF so little can be concluded about the generalisability of their findings.

As part of the assessment package for heart-lung transplantation, Whitehead et al (1991a), following extensive psychological evaluation by a psychiatrist and psychologist, with observation of family interaction, parental subsystem communication and problem solving, described 11 of their 22 families as functioning normally, 8 as having moderate dysfunction and 3 as being severely dysfunctional.

None of these interview and observational studies had control groups.

3.10.4 Family Coping

Several factors have been shown to contribute to better adaptation. These include father's involvement with the CF child and family (Allan et al, 1976; Boyle et al, 1976), mother being involved in activities outside the home (Allan et al, 1976), time made for joint family activities (Blumenthal, 1969) and a supportive relationship with the child's physician (Kulczycki et al, 1969; Blumenthal, 1969). Better adaptation has been found where parents use cognitive coping strategies of attributing meaning to a situation, ie finding an explanation such as in terms of religious beliefs or finding a scientific/medical explanation for the situation (Venters, 1981), sharing the burden with others (Venters, 1981) and practising denial (Cowen et al, 1985; Mattsson, 1972; Simmons et al, 1985), and using strategies to minimise the burden (Venters, 1981; Patterson, 1990). Patterson, (1990), found that getting involved in activities that enhanced self esteem, helped to manage tension or provided social support, appeared to benefit the CF child.

Hymovich and Baker (1985) asked CF parents what coping methods they used in times of difficulty. The most popular methods were praying, talking with someone and keeping busy but the authors do not comment on the efficacy of these techniques.

3.11 Summary

1. The salient features of aetiology, course of the illness, treatment and prognosis are described. The respiratory and nutritional complexities of CF require a major commitment from families as therapeutic resources. (3.1 to 3.3.4)

2. Studies using self report methodology have largely found no difference in levels of psychological disturbance between CF children and well comparison groups. Interview studies suggest that anxiety, particularly separation anxiety, may be more common in CF children. (3.4.1)

3. Self report studies of early adolescence mostly find no difference from well groups but some studies have found higher rates of anxiety and depressive symptoms in adult samples. There is a paucity of adequately described interview studies and the evidence here is equivocal. Self esteem in CF patients is robust. (3.4.2 to 3.4.5)

4. Most studies have found good social functioning in CF adolescents. Psychosexual development is delayed, paralleling delay in onset of puberty. Illness severity seems to be the main limiting factor in sexual behaviour. (3.6.1 to 3.6.3).

5. Educational achievements are possibly higher than would be expected on health grounds. Unemployment levels are higher than normal in CF adults although there is no evidence that anything

other than illness severity and difficulty in obtaining suitable work are the determining factors. (3.6.4)

6. Some but not all studies show raised levels of emotional disturbance, particularly depression, in mothers of CF patients. Mother's judgment of the severity of illness and her perception of stress is correlated with her level of mental health but is independent of objective measures of severity of illness. Most of the work in this area has been carried out on mothers of young children. (3.9.1)

7. Fathers are much less involved in the treatment of CF than mothers and show no difference from well groups in levels of emotional disturbance. (3.9.1)

8. Level of disability in the patient has no bearing on level of psychological adjustment of siblings. (3.9.3)

9. Marriages of CF parents are more likely to remain intact than would be expected in the normal population. Some couples report that CF puts the marriage under strain but others that it strengthens the marriage. (3.9.4)

10. The first year after diagnosis and the terminal stage of the illness are the most stressful periods for CF families. (3.10.1)

11. Early anecdotal accounts reported poor family functioning but lacked control groups. Self report studies of family functioning at the childhood and adolescent stage find that CF families are within the normal range. There are no interview or observational studies with control groups. (3.10.3)

12. A number of factors that lead to more satisfactory coping by families have been identified. (3.10.4)

CHAPTER 4: LITERATURE REVIEW - EXPRESSED EMOTION

4.1 Development of the Concept

The concept of Expressed Emotion (EE) evolved out of a series of epidemiological investigations which spanned 12 years at the MRC Social Psychiatry Unit, London, linking outcome in psychiatric patients to their social environment. In particular, in schizophrenic patients who returned from hospital to live with relatives, the re-emergence of florid symptoms was linked not only to the effective provision of medication but also to the amount of face to face contact with key relatives and the emotional climate of the home (Brown, 1985). Brown and Rutter (1966) and Leff and Vaughn (1976) devised a measurement technique for this emotional climate, the index of Expressed Emotion (EE) which ostensibly is an indicator of a relative's negative affect or intrusive overconcern towards the patient.

4.2 The Camberwell Family Interview

The traditional medium for eliciting Expressed Emotion ratings is the Camberwell Family Interview (Leff and Vaughn, 1985). This is a standardised semi-structured interview that takes between 60 and 90 minutes to administer. Key relatives in the patient's household are interviewed separately at crisis point, shortly after the patient's admission to hospital. All interviews are audiotaped for later analysis. The interview is designed to elicit 2 kinds of information: objective information concerning life in the household in the months leading up to a patient's admission and subjective information concerning the relative's attitudes and feelings towards the patient and illness. During the course of the interview, the emphasis shifts from symptomatic behaviour to more general patterns of family interaction. Although the interview procedure is governed by rigorous guidelines, the approach is a flexible one and the tone that of an informal conversation rather than an interrogation. The result is almost always a rich picture of family life and the quality of information obtained

in most instances provides quite a good idea of relatives' characteristic response in a time of crisis.

4.3 Ratings of Expressed Emotion

Raters listen not only to the content of speech but also to the way in which emotion is conveyed in the voice. These judgments may appear subjective yet, after a 2 week training course with rating of criterion tapes, people can be taught to agree on how to rate a particular interview. In all, 5 dimensions are rated: criticism, positive comments, hostility, overinvolvement and warmth. Criticism, hostility, and emotional overinvolvement only are used to determine the EE index.

Ratings are made both from reports of events in the past and from attitudes and behaviours displayed during the interview. A critical comment (CC) is defined as "a statement which, by the manner in which it is expressed, constitutes an unfavourable comment upon the behaviour or personality of the person to whom it refers". It can be evidenced in both the content of the statement but mainly by the tone of voice. Psychiatric patients often behave in odd or unusual ways when ill. A relative's mere recognition or description of characteristics of the patient, however unfavourable, is never of itself sufficient to define a comment as critical. For example, a comment like "he doesn't get up until noon" would only be counted as "critical" if said in a critical tone of voice. Hostility is a more forthright expression of negative sentiment which tends to criticise the person for what he is rather than what he does or is a general rejecting remark.

Emotional Overinvolvement (EOI) ratings are made from 3 sources: exaggerated behaviour - this can be exaggerated emotional response to events in the past or emotional display and dramatisation during the interview. Secondly, unusually self sacrificing and devoted behaviour is another indication of overinvolvement. Finally extremely overprotective

behaviour - usually age inappropriate ways of dealing with issues of autonomy and independence is evaluated.

4.4 Scoring EE

Criticism and positive remarks are scored with a frequency count. Hostility is rated on a categorical scale with scores from 0 - 3. Over-involvement and warmth are scored on a categorical scale from 0 - 5. Typically relatives are dichotomised into high and low EE using post hoc cut off points. A high EE household is one where one or more relatives has a high EE score. An interview is rated as high EE if there is one or more of the key features: high hostility, high EOI, high CC. Different studies have used different cut off points to define "high EE". "High EOI" has been variously defined as equal to or greater than score 3 or equal to or greater than 4. Any example of Hostility defines a relative as high EE. The cut-off for high CC has ranged from a count of 2 up to 10 critical comments. A cut off point of 6 critical comments has been the most commonly used definition in schizophrenic patients. Shifts in the cut-off point have the unfortunate effect that tabulated data often reflect optimised cut-off scores and therefore bias the overall predictive strength of the EE measure. As EE is scored as a continuous dimension, it is a pity that it is not evaluated more often as such.

4.5 What is Expressed Emotion?

Vaughn (1986) has given an interesting qualitative account of EE characteristics. Firstly she says that low EE relatives have respect for the patient's relationship needs which will vary between diagnoses. For example, low EE relatives of schizophrenic patients would be sensitive to requests for social distance or privacy but relatives of depressed patients would increase social support. Low EE and high EE relatives differ in their attitudes towards the legitimacy of the illness and the relative's level of expectations for patient functioning. Low EE relatives are prepared to

make allowances, show greater empathy and make more efforts to understand the patient's ordeal than do high EE relatives. Finally, the intensity of emotional reaction is different in the 2 groups. Undesirable behaviour will be described more objectively and dispassionately in low EE households. Low EE relatives do express dissatisfaction with the patient but it is muted. High EE relatives often respond as if the behaviour in question were a personal affront. Vaughn (1986) concludes that "the EE Index is best viewed as an indication of the emotional temperature in the household, a fluctuating meter of the intensity of a relative's emotional response at a given point in time".

Independent researchers have supported Vaughn's descriptions. Kuipers et al (1983) and Bertrando et al (1989) found that high EE individuals seem to talk more and listen less, to answer in an impulsive way during the CFI. MacCarthy et al (1986) found that highly critical relatives appear to provide a more complex home environment by responding to difficult behaviour in ways that made it harder for psychiatric patients to respond to the significant parts of their communication. Brewin et al (1990) found that critical and hostile relatives tended to make more attributions to factors personal to and controllable by the patients compared to low EE relatives. Hubschmid and Zemp (1989) reported that high EE was associated with less flexibility, more coercion and more vigilance. As a result, the home environment appeared to be more prone to conflict.

Criticism is associated with longstanding social impairment (Vaughn, 1986). Certain patterns of behaviour are more difficult for relatives to cope with including unpredictable behaviour (Greenley, 1986) and 'negative' symptoms. These observations affect an interaction between patient behaviour and relatives' ongoing response. Birchwood and Smith (1987) proposed that "EE includes a state component which is a thermometer of developing interactions between patient and relative". The EE effect is more noticeable in the more chronic patient populations. The effect of EE is weaker at the first episode compared to subsequent ones (Leff and Brown,

1977; Macmillan et al, 1986, Barrelet et al, 1990, Goldstein et al, 1989). Dare (1991) has suggested a possible link between the EE concepts and Minuchin's "psychosomatic family" concepts.

4.6 Replication of the Original Findings

There have been many prospective studies replicating the original finding that high Expressed Emotion is predictive of relapse in schizophrenia 9 to 12 months after discharge from hospital even when pre-morbid history, severity of symptoms and compliance with medication are taken into account. Two good reviews of this work have recently appeared (Parker and Hadzi-Pavlovic, 1990; Kavanagh, 1992). Parker and Hadzi-Pavlovic (1990) examined the aggregated data from 12 studies (908 subjects) and conclude that high EE is established as a significant predictor of relapse with an estimated odds ratio of 3.7. However, they also somewhat cynically point out that over the publication interval 1962-1988, the "EE effect" has been declining in the more recent publications. Longer term follow-up studies demonstrate that low EE households tend to delay relapse rather than prevent it (eg Hogarty et al, 1991). Kavanagh looked at 26 studies (1323 subjects) and found a median relapse rate over 9-12 months of 21% in low EE households and 48% in high EE households.

Not enough attention has been drawn to the fact that EE is a far more powerful predictor of relapse in men than it is in women (Hogarty, 1985; Vaughan et al, 1992). While some of this is a statistical artefact with smaller numbers of women in the sample leading to the statistical test having insufficient power for a small sample size, there could be important differences in the tolerance by relatives of behaviour they consider gender inappropriate (Goldstein and Kreisman, 1988). Several studies are of men only and many do not report results separately for men and women.

4.7 EE in Other Illnesses and Other Carers

While most of the research on EE has centred on schizophrenia, there is now some evidence that EE could be a more generalised risk factor for relapse in psychiatry and health psychology (Kavanagh, 1992). Vaughn and Leff (1976) found that if the spouse of a patient suffering from neurotic depression made 2 or more critical remarks on the CFI, this was predictive of relapse. This finding has been replicated. (Hooley et al, 1986a, 1989; Miklowitz et al, 1988). The effect has been found in bipolar illness (Priebe et al, 1989) and in success in maintaining weight loss (Fischmann-Havstad and Marston, 1984; Flanagan and Wagner, 1991).

The attitudes measured by EE are found not only in parents but in spouses (Hooley and Teasdale, 1989) and in carers in mental health hostels (Moore et al, 1992).

There has also been work in AN and BN (see 4.12).

4.8 Generalisation to Other Methods

If EE is indeed a robust measure of the emotional temperature of a household, then the EE effect should be demonstrable under conditions other than an interview with a key relative. It should also correlate with conceptually similar constructs.

Doane and colleagues (1981) developed a coding system (Affective Style) that classified audiotapes of a 10 minute family discussion of a problem salient to that household into "benign" or "negative" negative affective styles on the basis of 3 dimensions: benign criticism, harsh criticism and neutral intrusive (mind reading) statements. She found that negative profiles were associated with psychotic relapse. Using this methodology, Valone et al (1983), looking at a sample of distressed adolescents, Miklowitz et al (1984) and Strachan et al (1986), both studying schizophrenic patients, found an

association between negative Affective Style and EE measured after a parental interview similar to the CFI. Cook et al (1989) re-examined Valone's data and found that the negative interactions were bi-directional.

Hooley et al (1986) looked at the interaction patterns of 30 married couples where one partner was depressed. This was assessed by a videotape of a 15 minute discussion between spouses on a topic on which their opinions differed. It was then rated with the Kategoriensystem fur Partnerschaftliche Interaktion (KPI) (Hahlweg et al, 1984) which has positive, neutral and negative rating categories. The couples were also assessed for EE after a CFI. An association was found between criticism on the KPI and high EE, again lending support to the construct validity of EE.

Hahlweg et al (1984) found no relationship between CFI/EE and KPI (see above) seen in a sample of schizophrenic patients. However, the KPI was carried out 2 months after the Camberwell Family Interview after discharge from hospital. As the effect of time is confounded with assessment method, it is not possible to draw conclusions. The same problem arises in a study by Miklowitz et al (1989) who failed to find a link between CFI/EE and Affective Style measured 8 weeks later.

Berkowitz (1987) found that EE could be rated from video recordings of family interviews. Szmukler et al (1987) interviewed the parents of 20 anorexia nervosa inpatients using the traditional CFI/EE methodology. He then videotaped the same parents, their index child and siblings participating in a "picnic lunch" and rated these tapes for EE. He found a modest (Spearman's r =.25) correlation between EOI in the 2 settings and a much more powerful correlation between Critical Comment ratings in the two settings (r = .82). As Szmukler helpfully gives a scattergram of his data, it is possible to see that one outlier case (a high CC mother) must be making a considerable contribution to this high correlation but the relationship would just hold even without that case.

Le Grange et al (1992b) has used the medium of a family interview, the Standardized Family Interview (Kinston and Loader, 1984, 1986) and Vostanis et al (1992) the medium of a video recording of a systemic family therapy session.

Measuring Expressed Emotion with the Camberwell Family Interview and the Expressed Emotion Scale is undoubtedly an expensive and time consuming exercise. The interview (in its shortened form) takes 1 to 2 hours to complete. Raters need to attend a special 2 week (100 hours) training course and can spend 2 or 3 hours on rating each tape. So not surprisingly a search is on for a more convenient assessment tool.

There is now a growing literature on other ways of assessing EE attitudes that are as efficient but less time consuming (Kazarian, 1992; Kuipers, 1992). The best known of these is the Five Minute Speech Sample (FMSS) (Magaña et al, 1986) in which a key relative is asked to speak into a tape recorder for 5 minutes without prompting describing their thoughts and attitudes to the patient. This statement is rated for dimensions which are similar but not identical to EE ratings. The FMSS misclassifies 30% of high EE subjects as low EE and is unpopular with subjects (Jackson et al,1990).

In conclusion, EE seems robust enough a concept to be measured in mediums other than the original Camberwell Family Interview but alternative rating scales need to be treated with caution.

4.9 A Unitary Construct

There is some controversy as to whether emotional over-involvement (EOI), criticism and hostility reflect a single underlying construct or whether they have arbitrarily been brought together under the EE umbrella. The concepts of hostility and criticism may represent a degree of the same tendency towards fault finding, since in practice hostility is rarely found

without critical comments also being made. Many studies reporting some significant effect of EE in fact find this only on the critical comments dimension. The relationship between EOI and relapse did not reach significance in the early study of schizophrenic families by Brown et al (1972). In America, Hogarty et al (1986) and in Switzerland, Barrelet et al (1990) found that EOI in relatives did not predict relapse. EOI does not appear to be independent of pre-morbid functioning (Miklowitz et al, 1983; Vaughn et al, 1984). High EOI is only exceptionally present in spouses (Vaughn and Leff, 1976; Hooley et al, 1986) and therefore is of little predictive value in such households. In eating disorder populations high EOI did not predict dropping out of treatment (Szmukler et al, 1985; van Furth, 1991), nor treatment outcome (van Furth, 1991; le Grange et al, 1992a, 1992b).

4.10 Stability of EE Over Time

It is assumed that the EE rating reflects the type of family environment that the patient will encounter after discharge. EE is most evident on the CFI when assessed during a patient's admission. There is growing evidence that EE ratings reduce over time. Brown et al (1972) found that in about a third of cases the relatives' level of criticism dropped considerably following a marked improvement in the patient's condition after discharge. Many studies show that a substantial minority of high EE relatives become less critical over time (eg Hogarty et al, 1986; Tarrier et al, 1988; Leff et al, 1990). A small number of relatives become more critical. Therefore family interactions may change in quality over time.

There could however be an alternative explanation for this phenomenon. The CFI may be insufficiently sensitive to detect EE between relapses. There are real difficulties in carrying out repeats of the Camberwell Family Interview. The interview is long and although structured is informal and friendly in tone. The relatives are asked to "tell their story", to bare their souls and to allow the interviewer to intrude into their family life. Most

people are prepared to do this once. However, there is nothing naturalistic about asking people to repeat their story in this way a few months later, particularly if the interviewer is the same person as before. Follow up CFIs tend to be shorter and answers less elaborate than in the original CFI and relatives may be unwilling to show the same level of affect again. Reductions in EE with time have been observed in ratings made on videotapes of routine family therapy sessions between first and last therapy sessions (Vostanis et al, 1992). Hogarty et al (1986) found that 23% of the experimental group and 52% of the control group refused follow-up CFIs. Van Furth attempted to take 4 repeat measures of CFIs. By time 4, 33% of parents were refusing to do the CFI again. However, he reports that some families found the follow-up interviews helpful.

4.11 The Family Intervention Studies

The most important consequence of the recognition of the EE measure as an index of environmental stress has been to allow an empirical demonstration of the effect of the home environment on relapse rates. If the stressful family atmosphere could be modified, then patients might have fewer relapses.

To date 5 major intervention studies in schizophrenia have been published with 2 year outcomes (Leff et al, 1982, 1985; Falloon et al, 1982, 1985; Hogarty et al, 1986, 1991; Tarrier et al, 1988, 1989 and Leff et al, 1989, 1990). The treatment packages have involved family therapy, relatives groups, education, behavioural family intervention, social skills training and problem solving therapy. The aims of the interventions have been to prevent relapse and improve the patients' level of functioning and have been applied to high risk families. All 5 studies demonstrated the efficacy of psychological interventions in reducing relapse and, in the 4 studies where repeat EE measures were made, a change in relatives' EE from high to low was associated with lower relapse rates. The relapse rates at 9 months or 1 year in the family treatment group ranged from 6% to 23%

compared with 40% to 53% in the control groups. These effects could not be explained by differences in medication compliance, more energetic drug treatment or more contact with the clinical team. By 24 months the effect is less impressive as a fairly high proportion of patients has relapsed in most studies, 33% for family intervention, 71% for controls (Kavanagh, 1992). It looks as if the intervention packages serve to delay rather than to prevent relapse in the longer term. A recent Australian intervention trial using relatives' counselling groups rather than a behavioural package failed to influence relapse rates (Vaughan et al, 1992).

4.12 Expressed Emotion and Eating Disorders

It has already been mentioned that EE has been measured satisfactorily in mediums other than the CFI in eating disorder research (see 4.8).

Levels of Expressed Emotion appear to be considerably lower in eating disorder families than in schizophrenia families. Mean scores for critical comments in the schizophrenia studies have ranged from 2.4 to 8.2 compared to between 1.5 and 5.5 for eating disorder mothers and between 1.1 and 2.0 for eating disorder fathers (van Furth, 1991). Levels of emotional overinvolvement also appear to be lower in eating disorder families (Vaughn and Leff, 1976; le Grange et al, 1992a, 1992b). Rates of high EE are more similar to those of depressive outpatients (Hooley et al, 1986). Mean scores for eating disorder studies are summarised in Table 4.1.

TABLE 4.1 EXPRESSED EMOTION AND EATING DISORDERS

	n	Critical Comments Mean (SD)	EOI Mean (SD)
Szmukler et al (1985)			
AN and BN mothers	40	5.3 (4.8)	1.3 (1.6)
fathers	35	2.2 (2.1)	0.4 (1.0)
Szmukler et al (1987)			
AN mothers	20	5.5 (5.6)	1.1 (1.4)
fathers	20	2.1 (2.4)	0.2 (0.5)
van Furth (1991)[1]			
AN and BN mothers	24	2.2 (3.6)	2.0 (1.7)
fathers	22	1.7 (2.3)	1.1 (1.3)
le Grange et al (1992a)			
AN relatives	34	1.5 (1.9)	1.4 (0.8)
van Furth (1992)			
AN and BN mothers	47	2.6 (3.4)	1.8 (1.6)
fathers	47	1.9 (2.3)	1.0 (1.2)

[1] A subset of van Furth's 1992 results (see below)

In an interesting parallel with the schizophrenia family intervention studies, le Grange and colleagues (le Grange, 1989; le Grange et al, 1992a, 1992b), as part of the pilot stage of a study comparing 2 different forms of family work: conjoint family therapy versus family counselling in the families of AN patients, measured Expressed Emotion in 34 parents of 18 patients at base-line and at 6 months after treatment onset. They divided the sample into 2 groups - patients with a poor clinical outcome and those with good/intermediate outcome - and found that a high number of critical comments at baseline predicted poor outcome. They used a cut-off of 2 or more critical comments as a definition of "high". Furthermore, the families of patients with poor outcome appeared to become more critical as time went on. In the good outcome group, high EE reduced from 27% to 5% of the sample compared to an increase from 55% to 66% in the poor outcome group. Levels of Emotional Overinvolvement were initially low in both groups and did not change with time. No active attempt was made to reduce EE during the course of treatment.

Szmukler et al (1985) found that a high level of critical comments was a modest predictor of dropping out of treatment in the sample of AN and BN parents who were participating in a family therapy treatment trial (Russell et al, 1987). His group also found that the mothers and fathers of patients with BN were more likely to make critical comments than were parents of AN patients.

Van Furth (1991) has given an interim report on a series of Dutch families who are being monitored while their eating disordered child receives in-patient treatment. Half of the patients had AN. At the time of analysis, out of a possible 34, 26 families had EE ratings for intake and end of treatment. Van Furth found that an initial high number of critical comments in mothers was predictive of poor outcome but did not find this for fathers. However, he could not replicate le Grange's finding that change from high to low EE during treatment was connected with a better outcome. He could not account for this discrepancy by difference in methodology. EOI was of no value as a predictor in either study. Baseline data on van Furth's full sample are presented in Table 4.1 (van Furth, personal communication, 1992).

4.13 Summary

1. The EE concept, the Camberwell Family Interview and the rating system are described. (4.1 to 4.5)

2. Studies of the families of schizophrenic patients suggest that high EE is predictive of relapse with an odds ratio of 3.7. This effect has also been found in depression. (4.6, 4.7)

3. There is evidence for the validity of the EE concept in that it has been possible to measure it in settings other than the Camberwell Family Interview. It also has identity links with the Affective Style

concept and KPI criticism. Attempts to replace the EE/CFI procedure with briefer scales have met with limited success. (4.8)

4. There is evidence from schizophrenia studies that EE may change spontaneously over time. There is also evidence that therapeutic intervention designed to improve family functioning can also reduce EE levels in the families of schizophrenics. (4.9, 4.10)

5. Levels of EE appear to be lower in Eating Disorder families than in schizophrenia families. Whether high EE predicts dropping out of treatment remains equivocal, as does whether a change from high to low EE parallels a good treatment outcome. High levels of criticism in mothers at least seem predictive of a poor outcome to treatment. (4.12)

CHAPTER 5: METHOD

The broad plan was for the families of AN patients to be compared with the families of CF patients and a third group of well families. Assessment was to take place via 3 modalities: self report, interview and observation at baseline and at 8 months follow up.

5.1 Subjects

5.1.1 Sample Size

A power analysis was used to determine sample size (Pocock, 1983). Assuming a significance level of .05 and power of .90, an N of 22 subjects in each group would enable a difference between groups of about 10 points (equivalent to 1 SD) on the Family Assessment Measure to be detected. Allowance had to be made for some attrition between baseline assessment and the 8 month follow-up assessment. Without an extensive pilot stage it is difficult in an individual study to make an accurate prediction of drop out. Steinhausen's (1991) review of follow-up studies of anorexia nervosa suggests that between 11% and 24% are reasonable safety margins. A lot depends on the degree of effort made by researchers to pursue lost subjects. A target was set of recruiting 30 subjects for each group. During the recruitment period from September 1990 to October 1991, 27 subjects were recruited for the AN group, 29 for the CF group and 31 for the well group.

5.1.2 Inclusion criteria: All subjects

Certain inclusion criteria were common to all three groups: age range, geographical feasibility and "living at home".

The index child[1] had to be aged between 14 and 24 inclusive. This age band was a trade off between the need to select sufficient subjects and the need to keep the family stage relatively homogenous. Both anorexia nervosa and cystic fibrosis are rare conditions. The age band had to be sufficiently wide in order to recruit enough subjects. A standard reference source on family development (Duval and Miller, 1985) divides family development into 8 stages where different styles of family functioning are appropriate for different family stages. The age range 14 to 24 covers 2 of these stages: families with teenagers and families launching young adults, starting with the first child's departure from the home and ending when the last one goes. Although the 10 year age band enabled enough young people to be recruited, it did create difficulties in selecting appropriate instruments to measure family functioning and family burden. Families of older children in particular complained occasionally that they had "outgrown" the questionnaires.

As families were in the main to be interviewed at home, a distance limit was set at 2 hours travel time from hospital base. In the well group the distance limit was set at 45 minutes travelling time. The index child had to still be living in the parental home. Some of the index children were in the launching family stage. Leaving home is often a gradual process rather than a discrete event. "Living at home" was defined as spending at least 33% of their nights in the parental home or at least 25% of nights there and being in touch by telephone at least once a week. Young people "living at home" included several students away from home in term time but returning to parents every vacation and often during term as well. It excluded people who were married or co-habiting or who were single but whose domestic and social arrangements no longer had reference to their parents. Also included as "living at home" were 2 cystic fibrosis patients who lived alone in flats that were within walking distance of their parental homes and had their main meal daily with their parents.

[1] The term "child" is clearly inappropriate in some respects as the age range of subjects went from 14 to 24. Nevertheless it has been adopted both to mark the relationship to parents and as a convenient label for all subjects and not only those who were patients.

5.1.3 Inclusion Criteria: Anorexia Nervosa Families

In addition to the general inclusion criteria, the index child had to have met within the previous year the 4 DSM-III-R criteria for anorexia nervosa (Table 5.1). At the time of interview at least 3 of these criteria still had to be met. Thus the sample included both currently ill and partially recovered subjects. The onset of the illness had to have occurred at least a year prior to interview. This requirement was to mitigate the fact that CF families had had a long period of adaptation to illness.

TABLE 5.1 DSM-III-R CRITERIA FOR ANOREXIA NERVOSA

A. Refusal to maintain body weight over a minimal normal weight for age and height, e.g. weight loss leading to maintenance of body weight 15% below that expected, or failure to make expected weight gain during a period of growth leading to body weight 15% below that expected.

B. Intense fear of gaining weight or becoming fat even though underweight.

C. Disturbance in the way in which one's body weight, size or shape is experienced, e.g. the person claims to "feel fat" even when emaciated, or believes that one area of the body is "too fat" even when obviously underweight.

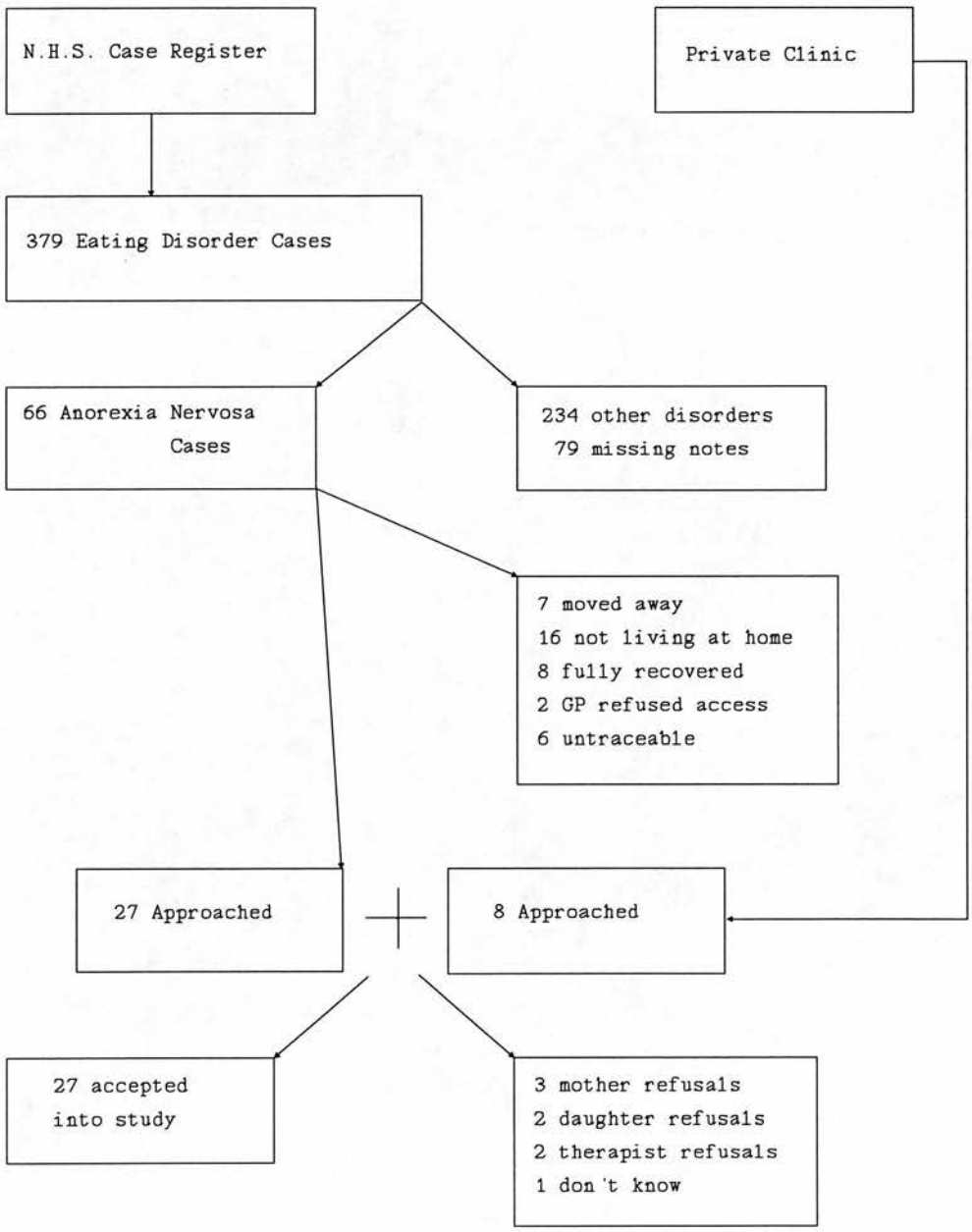
D. In females, absence of at least 3 consecutive menstrual cycles when otherwise expected to occur (primary or secondary amenorrhoea).

Of the total pool of 35 suitable families, 8 families (23%) refused to take part. Considerable efforts were made to establish the reasons for refusal. 3 mothers refused on their daughter's behalf. 2 of these said that their daughter was making good progress and did not wish to set things back by "dragging it all up again". The third mother said that the Clinic had always made her feel to blame for her daughter's illness and she could see no good reason to help. One girl said she was fed up with being included in research projects. One highly disturbed girl, battling against admission

to hospital and fighting with her parents, refused. 2 girls were encouraged to refuse by their therapist. The therapist concerned felt strongly that one of the major developmental tasks of anorexia nervosa was to move towards autonomy from parents and that interviewing the family would therefore be a retrograde step. One girl was unable to give a reason for refusal.

A flow chart of AN recruitment procedure is shown in Table 5.2.

TABLE 5.2
RECRUITMENT OF ANOREXIA NERVOSA PATIENTS AGED 14-24



5.1.4 Inclusion Criteria: Cystic Fibrosis Families

The index child had to be registered at a Regional Cystic Fibrosis Clinic which in all cases meant a clinically confirmed diagnosis of CF.

The reasons for selecting CF as the comparison group were in part theoretical and in part pragmatic. It was felt imperative to select a homogenous comparison group to simplify the number of variables that needed to be taken into account. The chosen illness needed to be one which could at least match AN in terms of severity or perceived threat of mortality. On these grounds, illnesses such as diabetes, skin disorders, coeliac disease were ruled out. Secondly, the potential recruitment pool needed to be of sufficient size. Physicians in oncology, rheumatoid arthritis and renal units were approached and, although interested in the project, could not guarantee sufficient numbers of patients. For example, in the Region there are fortunately only 6 new cases of cancer in adolescents in a year.

Cystic fibrosis was chosen because with advancing age it becomes a life threatening illness and has a clear cut non psychogenic aetiology. There is also the particular interest that weight can be low and the maintenance of adequate nutrition is of central importance in maintaining good health. Families are not passive bystanders but have a central role in managing the illness. There were a sufficient number of sufferers to include in the study. A potential disadvantage was that CF is a life long disorder whereas AN does not normally begin before the teenage years.

5.1.5 Inclusion Criteria: Well Families

Family members had to be "well". An operational definition of "well" was defined as follows. If a household had no one living at home who had attended a hospital within the previous 2 years or if no one attended their family doctor for a chronic condition, the family was defined as "well".

Typical examples of families excluded because of "a chronic condition" were a family with a child with multiple appointments for eczema attacks and a family with a grandmother with Alzheimer's disease living in the household.

5.2 Recruitment Procedures

5.2.1 Recruitment and Refusals - Anorexia Nervosa Families

Anorexia nervosa subjects were recruited from 2 sources: NHS medical records and a private clinic.

A computer search was carried out for all patients diagnosed as "Eating Disorder" currently in the appropriate age band and referred since 1984 to the psychiatric services of a Scottish Regional Health Authority (total population 743,700). This produced 379 cases of which alarmingly 79 sets of casenotes were missing from medical records. Examination of the casenotes permitted a confident diagnosis of anorexia nervosa to be made on 66 cases. Next the current therapist or family doctor of these patients was contacted for up to date information and permission was sought to contact the patient to invite them into the study. At this point a further 39 cases failed to meet the entry criteria: 7 patients had moved away from the region, 16 were not living at home, 8 were fully recovered. In 2 cases the family doctor refused access. We were unable to trace 6 cases in spite of energetic efforts to do so: 3 no longer appeared to be registered with a family doctor and 3 were not at their last known address.

Patients were also recruited from a private psychiatric clinic with a mixed patient population that has a special interest in eating disorder patients. 8 suitable cases were identified during the study recruitment period.

The identified suitable cases were invited into the study as follows. The index child was sent a letter explaining the objectives of the study and the research procedure. If under 16, the letter was sent jointly to parents and

child. He/she was asked to discuss the letter with the parent(s) and told to expect a telephone call at home in a few days time when the family would be invited into the study. In-patients, of whom 7 were recruited, were either telephoned in hospital or received a personal visit.

5.2.2 Recruitment and Refusals - Cystic Fibrosis Families

CF patients and their families were recruited from 2 specialist regional centres for adult patients and one paediatric specialist unit. These clinics had a catchment area of the central belt of Scotland (central belt population 3,000,000). It has been convincingly shown that life expectancy is better when treatment is given at specialised clinics (Nielson and Schiotz, 1982; Warwick, 1982) and so present health policy is that all CF patients should be registered with such a clinic by their general practitioners. The usual age of transfer from paediatric to adult service is about 14, account always being taken of both the social maturity of the young person and practical considerations such as travelling distance. The paediatric clinic involved acted as a feeder clinic for one of the adult clinics. However, for historical reasons, the paediatric unit had 3 young people of age 19, 20 and 20 still attending. All potential recruitment sources for CF patients in the 14 to 24 age cohort in the central belt of Scotland were tapped.

Approaches to the families of CF patients differed slightly between the 2 adult centres. This was to accommodate the different organisation patterns of the 2 clinics and the wishes of the consulting physicians. Adult Centre A held CF clinics which were attended by patients with routine appointments. Clinic lists were available a fortnight ahead and all patients in the appropriate age band still living at home were sent a letter explaining the objectives of the study and informing them that they would be approached by a research worker at their next clinic visit to invite them into the study. All clinics except one were attended by a research worker over the recruitment period October 1990 to April 1991. 20 patients had a personal approach from a research worker. Of these, 3 families refused.

Reasons for refusal were that one patient's parents were in process of divorcing, the mother of a second patient said that an interview would be too distressing for her and the father of a third patient vetoed his family from participating because "he had no time for psychology". 10 further potentially suitable recruits were not approached by the research worker either because they did not attend (non-attendance ran at 30% at this clinic), because they were well enough to be very infrequent attenders or because they by-passed the monthly clinic through an emergency appointment or admission to the ward. Thus these patients ranged from very well to very ill. As the target number of patients was reached, there was no need to pursue this group.

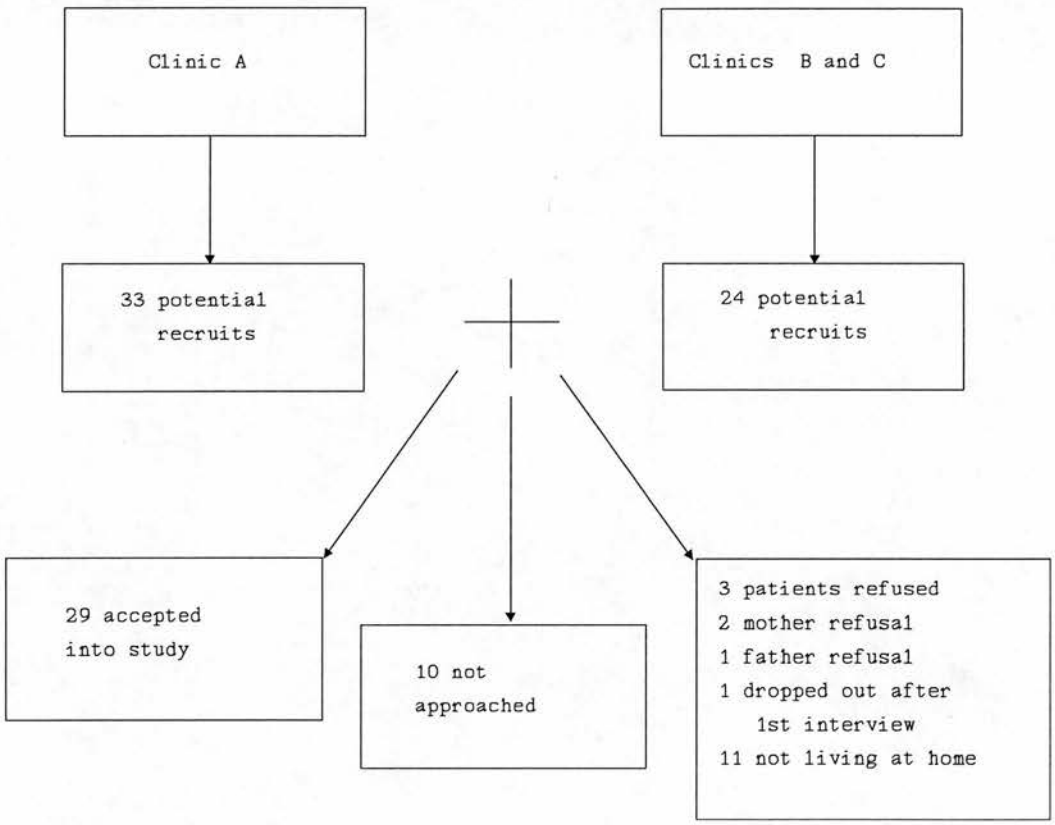
Adult Clinic B had much smaller weekly clinics, usually with 1 to 4 attenders. Approaches to patients were all made through a liaison doctor. She gave an information sheet to each patient who met the research criteria on routine visits and then arranged for the research worker to telephone the patient direct. One patient was not approached as the doctor felt it would be too distressing for the family, a twin who also suffered from CF having recently died. All other suitable patients registered with this clinic were approached.

3 families from Clinic B refused. A patient in good health with no known psychological difficulties or family problems wrote to say that she did not wish to take part. A girl of 17 agreed to take part but her mother later wrote to say that she wished her family affairs to remain private. The family of this girl were in fact unknown to the clinic, a most unusual situation. A young man who had 2 years earlier been referred to a child psychologist for oppositional behaviour was the third refuser. One patient could not be included as his brother, also a CF sufferer, had already come into the study. A further family dropped out after the first interview.

The liaison doctor also covered the paediatric CF clinic (Clinic C) and the 3 adults attending this clinic were all recruited in the same manner.

The refusal rate for the CF sample was 16%. There was no difference in refusal rate between the 2 main centres. A flow chart of CF recruitment procedure is shown in Table 5.3.

TABLE 5.3
RECRUITMENT OF CYSTIC FIBROSIS PATIENTS AGED 14-24



5.2.3 Recruitment and Refusals - Well Families

2 methods of recruiting "well families" were used: nomination by ill group families and selection from GP clinics.

If a CF or AN family lived within 45 minutes of the research centre, the family were asked to nominate a "well" family as defined above. 6 families were recruited from the AN sample and 9 from the CF sample in this way. After permission had been obtained to approach nominated families, these families received an information sheet inviting them into the study explaining the objectives of the study and telling them to expect a telephone call in a few days time. 2 families approached by this method refused to be interviewed saying that "it was not convenient".

The remainder of the well families were recruited from 2 large urban GP practices, one with a middle class and one with a working class catchment area. Computer print-outs of all girls in the 14 to 24 age cohort still living in families were produced. Casenotes for each member of a household were examined in alphabetical order until the recruitment target was reached. 25% of girls were not living in parental families. At the middle class practice this was usually because they were students at college or university away from home. At the working class practice this was usually because they were mothers. Of the girls still living at home, 30% of families were rejected because they did not meet the health criterion. Suitable families were approached by a letter from the research worker which explained why they had been selected, explaining the objective of the study and told them to expect a telephone call. 2 families refused, with no reason being given.

The refusal rate for well families was 11%.

5.3 Baseline Assessment

Family assessments were made at 2 stages: baseline and at 8 months follow-up.

Baseline information for the AN and CF families was collected in 2 interviews. Interview 1, carried out by an independent assessor[1] blind to the hypotheses of the study, was with the index child alone. At this interview the health status of the index child was established, demographic information obtained, family questionnaires completed and information elicited to assist in the selection of salient family problems for the Problem Solving Task. This interview lasted approximately three quarters of an hour. These measures are described in detail below. This interview was usually held in the appropriate clinic.

Interview 2 was carried out a few days later by the author. 85% of interviews were carried out in the parental home at a time and place convenient to the family. In most cases this was in the evening. This step was taken to ensure as high a compliance as possible of all members of the family triad (mother, father and index child) and to provide an environment in which family members would feel relaxed. At this second interview, parental self report questionnaires were administered and parents were interviewed together using the Camberwell Family Interview. Then the index child joined his/her parents and the Problem Solving Task took place. This session lasted between one and a half and 2 hours. The measures are described in detail below (5.4 to 5.7.6) and summarised in Table 5.3.

The procedure for the well families was slightly different. Constraints of time and resources did not make it possible for the independent assessor to carry out Interview 1. Interview 2 followed on immediately from Interview 1 and both interviews were carried out by the author.

[1] Mrs Wilma Warwick, Research Associate, University of Edinburgh

TABLE 5.3 SUMMARY OF ASSESSMENT TOOLS: BASELINE ASSESSMENT

Interview 1

(Index Child)

Demographic Data

DSM-III-R interview for anorexia nervosa, bulimia nervosa and major depressive episode

Morgan and Russell Scales

Eating Disorder Examination (selected items)

General Health Questionnaire

Family Assessment Measure

Edinburgh Family Scale

Family Problem Interview (Doane)

Interview 2

(Parents Together)

General Health Questionnaire

Family Assessment Measure

Edinburgh Family Scale

Impact on Family Scale

Life Events Scale

Camberwell Family Interview

(Family Triad)

Problem Solving Discussion

5.4 Measures of Health Status of the Index Child

The following measures were given to all index children:

5.4.1 Morgan and Russell Scales

The Morgan-Russell outcome assessment schedule (Morgan and Hayward, 1988; Morgan and Russell, 1975) has become widely used for the clinical assessment of anorexia nervosa (Morgan and Russell, 1975; Morgan et al, 1983; Hall et al, 1984; Russell et al, 1987; Channon et al, 1989; Crisp et al, 1991; Ratnasuriya et al, 1991; Van Furth , 1991). The assessment takes the form of a guided interview which is concerned with clinical features central to the syndrome of anorexia nervosa. It evaluates eating behaviour, body weight, mental state, menstrual function, sexual and socio-economic adjustment. The responses to specific questions are scored on a 12 point scale with anchor points based on a combination of assessment at interview and clinical status. Information should preferably be obtained from both the patient and a key informant such as a parent.

The assessment produces 14 scaled ratings which are reduced to 5 scores by averaging subscale scores. The ratings can be averaged further to produce one single measure, the Average Outcome Score.

In the original version, assessments and scores were based on the 6 month period preceding the interview. As the follow-up period in the present study was only 8 months, it was decided that, in order to have a stronger definition between health status at Time 1 and Time 2, the assessment period should be restricted to one month before the interview. Menstrual status, however, was measured over a 3 months period. A further modification was that one of the 5 items on the Socioeconomic subscale, "relationship with nuclear family", was excluded from all analyses of the Morgan and Russell Scale. The reason for this is that the item is obviously confounded with family functioning which for the objectives of this study

needed to be a variable distinct from severity of illness. The "Socioeconomic" subscale mean therefore was calculated from the 4 remaining items.

5.4.2 Eating Disorder Examination Items and DSM-III-R

The Eating Disorder Examination (EDE) (Cooper and Fairbairn, 1987; Cooper et al, 1989) is a semi-structured interview measure focusing on current levels of specific psychopathology of anorexia nervosa and bulimia nervosa, particularly attitudes to weight and shape. It was decided not to use the scale in full for 2 reasons. Firstly, many items overlap with the Morgan and Russell scales. Secondly, it takes one hour to administer. For this study selected items only were administered: those that could generate operationally defined DSM-III-R diagnoses for anorexia nervosa and bulimia nervosa and items to assess bulimia, vomiting and exercise patterns which are not included in the Morgan and Russell Scales. Items from Version 11.3D of the scale were used.

5.4.3 Body weight

Weight was measured both by average body weight and Body Mass Index.

Heights and weights of index child were collected for all 3 groups. One of the diagnostic criteria for anorexia nervosa (DSM-III-R) is that there should be weight loss leading to maintenance of body weight 15% below that expected for age and height. There is unfortunately no consensus on which weight tables should be used in eating disorder research. This is not a trivial matter as Pyle et al (1986) have shown that the percentage of a sample of eating disordered patients classified as "underweight" ranged from 16% to 71% depending on which of 3 widely used weight tables were used. A further difficulty is that no one generally available table satisfactorily covers both the adolescent and young adult population covered by this study. In Britain the most commonly used tables for the under 18

age group are the Tanner and Whitehouse Charts (1966). However, at the extremes of height, estimates of expected weight differ alarmingly (by up to 10 kg) from, for example, the weight estimates for adults of the North American Build Study (1979) that provide the widely used tables of the Metropolitan Life Assurance Company. In order to have one set of weight tables only, expected weight from height and age was calculated from linear regression co-efficients from the DHSS height and weight survey for Great Britain (Knight, 1984).

As an alternative to describing weight as a percentage of average body weight for age and height, various indices have been constructed to measure relative weight. The most satisfactory of these is the Body Mass Index (BMI), sometimes known as the Quetelet Index. This is a ratio W/H^2 where weight is measured in kilograms and height in square metres. Median value of BMI in Britain in the 16 to 24 age group is 22 for both men and women (Knight, 1984). A commonly used classification (Llewellyn-Jones and Abraham, 1984) defines a BMI of less than 19 as "underweight" and less than 15 as "emaciated". Beumont et al (1988) suggest that a cut-off of BMI of less than 16 could be used as a diagnostic criterion of anorexic weight. However, great care does need to be taken in interpreting a BMI score when it is born in mind that the percentage distribution of "underweight" decreases sharply with age: 33% of 16 to 19 year olds have a BMI of 20 or less but only 7% of 40 to 44 year olds (Knight, 1984).

5.4.4 General Health Questionnaire

The General Health Questionnaire (Version GHQ30) is a self-administered screening test designed for use in psychiatric disorders in community settings and non-psychiatric clinical settings. It is aimed at detecting those forms of psychiatric disorder which may have relevance to a patient's presence in a medical clinic so that its focus is on psychological components of ill health. It has been used in a wide variety of settings as a screening

tool for psychological disorder and has been validated against standard psychiatric interviews (Goldberg, 1987).

5.4.5 Mental Health History

It is known to be extremely difficult to obtain reliable retrospective information sufficient to make particular psychiatric diagnoses. Instead a more global assessment was made of absence or presence of "mental health history". A "mental health history" was defined as any psychological disorder (other than the index illness) for which the subject had received treatment from the family doctor or a mental health professional. In addition, current DSM-III-R ratings of "major depressive illness" were made using operationalised DSM-III-R criteria.

5.5 Additional Measures for Cystic Fibrosis Patients

5.5.1 Cooperman Scale

The health status of all CF patients was assessed at the start of the project by their chest physician using the Cooperman Scale (Cooperman, 1971). This is a clinical evaluation covering 5 dimensions: general activity, chest radiographic finding, degree of clubbing, growth and development, and complications. In the style of the Apgar system, each dimension is scored 0, 1 or 2. There are anchor points for each score. The scores are summed to provide a total score. A patient in excellent health could have a maximum score of 10. A copy of the scale is found in Appendix VIII.

5.5.2 Lung Function Tests

Pulmonary disease is the single most important factor in both the morbidity and mortality of CF. All patients with pulmonary disease have progressive airflow obstruction. Two measures of lung function were carried out by clinic physiotherapists using a Vitalograph instrument. Forced expiratory

volume in one second (FEV_1) measures the volume expired during the first second of a forced expiration. Forced vital capacity (FVC) is the total volume of air expelled during forced maximal expiration following maximal inspiration. In the normal population, both these functions increase in relation to height, weight and age until the age of about 25. The results are customarily expressed as a percentage of the predicted normal value for height, weight and age. The FEV_1 is normally 70% or more of the FVC and this ratio falls when there is airway obstruction. Results should be considered abnormal if they are more than 20% below predicted value (Goodchild et al, 1989).

5.6 Health Status of Other Family Members

5.6.1 GHQ

General Health Questionnaires (GHQ30) were given to all family members of the household aged 13 and over.

5.6.2 Mental Health History

Mental health histories were taken of parents using the same criterion of "mental health history" as with the index child.

5.7 Family Functioning

There are 2 broad approaches to measuring family functioning. Firstly, there are instruments based on the reports of family members. Secondly, there are instruments based on the direct observation of some type of family interaction. Self report procedures include both paper and pencil tests and structured interviews. They have the advantage of being convenient and relatively inexpensive and allow for the possibility of collecting data from large representative samples to which individual test results can be related. They can capture family members' thoughts and attributions about

relationships and events which can be important predictions of family processes and outcomes. However, the individual's perceptions of self and others can be biased or inaccurate and can distort the views of what other observers might say about the same events. Observational procedures can range from laboratory analogues, for example observation of family performance of a structured task or game, to naturalistic observation in the home. Observational procedures inform us most directly about actual interchange among family members although the reactivity of the observer will always rule out a completely naturalistic event. At the same time they are expensive and labour intensive and thus large normative samples are not available. They present major but not insuperable difficulties in achieving clinically relevant coding systems or rating scales with good reliability (Eisler, 1985; Jacob and Tennenbaum, 1988).

In an attempt to meet some of the shortcomings of both these approaches, it was decided that a multi-method assessment of family functioning was appropriate.

5.7.1 The Family Assessment Measure

The Family Assessment Measure (FAM) (Skinner et al, 1983) was given to all family members of the household aged 13 and over. (Skinner developed the test for people aged 13 and over). As norms for this scale were not available for British subjects, it was also given to 214 schoolchildren and their families in the schools project test battery. Full details on the reasons for selecting this particular scale and information on its properties can be found in Chapter 9: The Schools Project (Section 9.5). A copy of the scale is in Appendix VII.

5.7.2 The Edinburgh Family Scale

This scale, developed for use in this project, as part of the schools test battery, was given to all family members of the household aged 13 and over. Full details of the development of the scale can be found in Section 9.6 and a copy of the scale in Appendix V.

5.7.3 Expressed Emotion Ratings

A modification of the Camberwell Family Interview (CFI) (Leff and Vaughn, 1985) was used as the medium for eliciting Expressed Emotion. The interview was shortened so that instead of lasting between 1 and 2 hours, it lasted between 45 minutes and 1 hour. 2 sections were cut. One, the items on "nagging" as it was felt that this area was adequately covered by the sections on "irritability" and "quarrelling". The second cut was a section discussing in detail the marital relationship. The present health status section was originally designed to elicit information on schizophrenic behaviour. Items were substituted so that typical AN and CF behaviour was covered. For example, AN parents were asked to describe the daily eating pattern of their child and were asked to describe exercise patterns. CF parents were also asked about eating patterns and were asked about compliance with the physiotherapy regime. Further, modifications had to be made to the interview for the well families. In the opening section of the interview relatives are asked to describe in detail health status in recent months. This was clearly inappropriate for well children. Instead parents were given the opportunity to discuss any episode of ill health and any episode in the child's history when parenting had been a particular challenge. Expressed Emotion is thought to be independent of the severity of health status so that a parent could theoretically obtain a high emotional overinvolvement rating when discussing, for example, an objectively mild bout of 'flu . Copies of the interview schedules can be found in Appendices I and II.

Parents were interviewed together, not separately as in the original CFI studies (Leff and Vaughn, 1985). EE ratings have successfully been made in media other than the CFI (le Grange,1989; Szmukler,1987).

Interviews were carried out by the author with both parents present and were recorded on audiotape. They were later rated by the author for Expressed Emotion. The author had trained as an EE rater on a standard 2 week course in February 1990. Trainees are taught to rate a set of standard criterion tapes to satisfactory reliability levels.

An independent assessor[2], blind to the objectives of the study but experienced in rating AN families, rated a sample of 15 tapes, 5 from each patient group, 29 parents in all (Table 5.4). The mean difference between raters was 0.17 critical comments with 95% limits of agreement ranging from -1.23 to +1.29 critical comments between raters which seems clinically acceptable. There was a mean difference of 0.27 positive remarks but this time the 95% limits of agreement were wider (-3.52 to +2.96).

As Emotional Overinvolvement (EOI), Hostility and Warmth are rated categorically the appropriate measure of agreement is the weighted kappa statistic (Altman, 1991). Agreement between raters for EOI was very good (k = .84) but only moderate for warmth (k = 0.53). No ratings of hostility on these tapes was made by either rater, thus giving perfect agreement. (See 5.8 for a discussion of the "limits of agreement" approach and the kappa statistic).

[2] Dr D le Grange, Institute of Psychiatry, London.

TABLE 5.4 EXPRESSED EMOTION - AGREEMENT BETWEEN RATERS

n = 29 Parents			
Dimension	Weighted Kappa Statistic	95% Confidence Interval	Strength of Agreement
EOI	0.84	.61 to 1.0	Very good
Warmth	0.53	0.73 to 0.33	Moderate
Hostility	1.00		Perfect
	Mean difference	95% limits of agreement	
Critical Comments	0.17	-1.23 to +1.29	
Positive Remarks	0.27	-3.52 to 2.96	

5.7.4 The Impact-on-Family Scale

The Impact-on-Family Scale (Stein and Riessman, 1980) is a 24 item self-report scale designed to elicit the impact of a child's illness on the family system. It has 4 subscales: financial impact, familial and social impact, personal strain and mastery, which are measured on a 4 point Likert scale ranging from Strongly Agree to Strongly Disagree. Internal consistency for the scale ranged from .86 to .60. The scales can be summed to form a total score.

Minor modifications were made to the (1978) version of the scale to make it more appropriate to the study population. Item i was omitted as being age inappropriate. Item m was omitted as it was ambiguous. On item c "I" was changed to "I/my partner". On item aa "acutely ill" was changed to "very unwell". Item z was shifted to the Personal Strain factor as it correlated better with that subscale than with the Mastery subscale. As in the 1980 version of the scale, items e and s were omitted (see Appendix VI for a copy of the scale).

Mothers and fathers were asked to complete the scale.

5.7.5 Life Events Scale

This 22 item scale was a modification of the Holmes and Rahe (1967) schedule developed in the schools battery for use on an adolescent population. (Full details of the development of the scale are given in Chapter 9: Section 9.7 and a copy of the scale is in Appendix IV).

5.7.6 The Problem Solving Task

The objective of the problem solving task was for the family triad of mother, father and index child to discuss an issue or problem salient to their household and for this interaction to be assessed for presence of some of the concepts that Minuchin measured in his original work. Each family discussed 2 problems lasting up to 10 minutes. The discussions were audiotaped, transcribed and rated for mind reading, mediating response, alliance shifts, closeness, overprotectiveness, weak generation boundaries and resolution of conflict.

Final scores were obtained for 3 concepts: (1) "enmeshment/ overprotection" which was the sum of alliance shifts, mind reading, mediating response, closeness and overprotection. Scores could range from 0 to 20 (2) "generation boundaries". Scores ranged from 0 to 4. (3) "Problem solving skills" were scored in 3 ways. Families were given a classification of "good", "medium" or "poor" problem solving skills depending on whether they obtained a satisfactory solution on both, one or neither of the 2 problems under discussion. They were also rated as to whether they used conflict diffusion or avoidance as a discussion technique on a 3 point scale "no diffusion", "some diffusion" or "clear diffusion". Finally they were rated as to whether open conflict was used as a technique or not: "no open conflict", "some open conflict", "high open conflict".

Full details of the development of the Problem Solving Task and its scoring system are found in Appendix III.

5.8 Statistical Note

Statistical analyses were carried out by myself on a PC using modules from the statistical package CSS: STATISTICA. The only exception to this were calculations of weighted kappa which is the statistic of choice to measure agreement between raters for ordered categorical data. This is not available on any of the standard statistical packages and so was painstakingly calculated by hand in the medical statistics unit[3].

It is only in recent years that I have come to appreciate that medical statistics are as much art as science. For example, individual statisticians are likely to have different views on how far a particular data set has to stray from a normal distribution before it becomes relegated to the 2nd XI of non-parametric methods. I have aligned myself to the advice given by one particular statistician, D G Altman, in his recent excellent textbook. Together with the late M J Gardner, he has guided recent statistical policy in the British Medical Journal and the Lancet (Gardner and Altman, 1989; Altman, 1991).

5.8.1 Estimation and Hypothesis Testing

Altman (1991) is a persuasive advocate for the estimation (confidence interval) rather than hypothesis testing (p values) approach to data analysis. It seems intuitively sound and has the great merit that it forces the reader to look at the data and make a clinical judgment on whether the results are "good enough". However, confidence intervals are only rarely reported in this study for the pragmatic reason that routines are not widely available on PC statistical packages and need to be calculated by hand.

Where possible exact p values are reported although occasionally the $p < 0.05$, < 0.01 and < 0.001 convention is followed. In all cases 2 tailed tests are used. The term "significant" is reserved for results where $p < 0.05$.

[3] Mr Jian-Hua Mao, University of Edinburgh

In the tables p values less than .05 are printed in bold. NS means not significant, ie $p > .05$.

5.8.2 Continuous Data

For continuous data differences between the 3 groups were tested with one way analysis of variance (ANOVA). The normality assumption of the test was checked with the Kolmogorov-Smirnov statistic and the homogeneity of variance assumption with Bartlett's test. Where the data failed these tests, the non-parametric one way analysis of variance Kruskal-Wallis test was used. In order to reduce the chance of Type I errors, where the F ratio was significant, post hoc paired comparisons were made using Duncan's Multiple Range test rather than a simple t test. No such correction exists for non-parametric data so here the Mann Whitney test was used between pairs.

Again to avoid Type I errors, where there were multiple dependent measures that were likely to intercorrelate such as the subscales of the FAM test, a multivariate analysis of variance (MANOVA) was carried out. If the test statistic (Wilks' lambda) was significant, then univariate F tests were carried out for each variable.

The follow up study necessitated the use of some repeat measure statistics. The 2 way MANOVA design selected to measure change in the self report family functioning scores is described in detail in 7.7.1. Again the rationale was to limit Type I errors. At follow up, there were several comparisons between the AN and CF groups only. Here the paired t test was used and the Wilcoxson's matched pairs test for non-parametric data.

5.8.3 Categorical Data

The χ^2 test was used to test for significant differences between proportions such as the proportion of psychiatric "cases" on the GHQ test. The Median test simply splits data into 2 categories at the median of the total sample so that it can then be analyzed using χ^2. This approach was used to test for differences between groups on the 3 household climate variables (see Chapter 6.8). In the follow up study, differences between baseline and follow up scores were tested with McNemara's χ^2 for matched proportions.

5.8.4 Correlational Data

It should be assumed that Pearson's r statistic was used throughout unless (as will be stated in the text) the data required the non-parametric Spearman's r statistic to be used.

5.8.5 Multivariate Data

Principal components analysis was used to derive 3 'household climate' variables from 7 correlated variables (see 6.8).

5.8.6 Agreement between Raters

At several points in the study it was necessary to establish whether 2 raters could agree (5.7.3, 7.6.1, Appendix III). For categorical data kappa and weighted kappa were used. Weighted kappa is appropriate where categories are ordered. Altman (1991) suggests the following guidelines in interpreting kappa. Values < 0.2 poor; 0.21 to 0.40 fair; 0.41 to 0.60 moderate; 0.61 to 0.80 good and 0.81 to 1.00 very good. For continuous data many studies give a correlation coefficient as an indicator of agreement between raters. Altman and colleagues (Bland and Altman, 1986; Altman, 1991) argue convincingly against that approach and suggest that the point of interest is the mean difference between raters and the range of that

agreement for individuals (using confidence intervals), the "limits of agreement" approach which was used here.

5.8.7 The Schools Project

Development of the Edinburgh Family Scale was closely modelled on procedures suggested by Nunnally (1978) and Kline (1986) using item analysis and principal components analysis. The methodology is described in the text (Chapter 9).

5.9 Main Hypotheses of the Study

The general objectives of the study are set out in the Introduction. The specific hypotheses to be tested are as follows:

1. If AN families are more dysfunctional than other families with illness, then they should show higher Family Assessment Measure (FAM) and Edinburgh Family Scale (EFS) scores than the CF and well groups. They should show more Expressed Emotion and more difficulties on the Problem Solving Task.

2. If Minuchin's description of the "psychosomatic family" is correct, then the following predictions would be made.

(a) On the FAM scale, the AN should show significantly lower scores on the Involvement and Affective Expression subscales than the CF families who in turn should show lower scores than the well families. They should show higher scores on the Task Accomplishment subscale.

(b) AN families should show higher (more dysfunctional) scores on the Edinburgh Family Scale (EFS) subtests than the CF families who should have higher scores than the well families.

(c) AN families should have higher (more dysfunctional) scores than the CF families who should have higher scores than the well families on Conflict Resolution, Enmeshment and Overprotection on the Problem Solving task.

(d) AN families should show more Overinvolvement as defined by Expressed Emotion ratings than the CF families who should show more Overinvolvement than the well families.

(e) It follows from Minuchin's conceptualisation that between the first assessment and the 8 month assessment, in AN families, increase in family dysfunction should correlate with an increase in illness severity. Any similar correlation found in cystic fibrosis families should be significantly weaker.

If significant differences are indeed found between the AN families and the other groups, alternative explanations for those differences need to be ruled out.

(a) **Affective Illness** Differences between family groups should hold when GHQ scores of family members are held constant.

(b) **Life Events** There is widespread interest in the effect of significant life events on illness (eg Cleary, 1980). Differences between groups should still hold when number and significance of life events is held constant (Holmes and Rahe, 1967).

(c) **Burden of Illness** If poor family function is not just the result of the burden of illness, then differences between groups should hold when illness burden (Impact on Family Scale) is held constant.

CHAPTER 6: RESULTS

6.1 Demographic Characteristics of Families

The demographic characteristics of the families are summarised in Table 6.1.

6.1.1 Index Child

There was no significant difference between groups for mean age. However the groups were not matched for sex. As expected, the majority of the anorexia nervosa patients were female (93%) but unexpectedly only 32% of the cystic patients were female. For reasons which are not understood, survival rates for this illness are higher in males (Goodfellow, 1989). However, this factor alone cannot account for the low ratio of women. The ratio was a true reflection of those patients registered at the CF clinics so that distortions in the response rate cannot account for this difference either. There was a significant difference in the work status of the index child (χ^2 = 20.99, p = .002). While all of the well children were either working or school and college students, some 19% of the AN group and 41% of the CF group were unemployed. All in the ill groups were unemployed for factors related to their illness. Half of the CF group were genuinely unfit for work through illness but the other half were caught in a social security trap, being fit for part-time or light non-manual work but finding that their financial position was worse if in work than if benefit dependent. This problem was particularly acute for those coming from working class households.

6.1.2 Mothers

Mean age of the mothers was 45.2 (SD 6.6) with the ill family mothers being a little older than the well family mothers. 75% of mothers were working either full or part-time with no significant difference between

groups. There was no relationship between whether a mother worked and the severity of illness of her child in either of the ill groups. CF mothers were encouraged by the physicians to seek interests outside the house and most actively pursued this policy.

6.1.3 Fathers

Mean age of fathers was 48.0 (SD 6.4) with no significant difference between groups. 97% of the fathers were working with 4 retired and 3 unemployed.

TABLE 6.1 AGE, SEX AND EMPLOYMENT OF FAMILIES

	Anorexics	Cystics	Well
Index Child	n = 27	n = 29	n = 31
Age Mean (SD)	18.7 (3.2)	19.2 (2.7)	18.2 (2.6)
Male:Female	2:25	20:9	8:23
Work Status			
Employed	15%	24%	32%
Student	66%	35%	67%
Unemployed	19%	41%	0%
Mothers	n = 27	n = 29	n = 31
Age Mean (SD)	47.4 (5.2)	45.9 (6.4)	42.7 (7.1)
Work Status			
Full time	31%	28%	35%
Part time	35%	45%	52%
Unemployed	35%	28%	13%
Fathers	n = 23	n = 25	n = 29
Age Mean (SD)	50.0 (6.9)	48.4 (6.7)	46.2 (5.3)
Work Status			
Employed	86%	89%	97%
Retired/Unemployed	14%	11%	3%

6.1.4 Family Structure

Family structure is summarised in Table 6.2.

In every case the index child was living with his or her mother. 89% of these mothers were married with no significant difference between groups for marital status. In 3 of the families mother's husband was a step-father to the index child but was treated as father in all analyses. Conversely the 6 fathers (2 in each group) who were divorced or separated from their wives were not contacted for interview as they were no longer living in the parental home.

The mean number of children in the family was 2.3 (SD 0.91) with no significant difference between groups.

Some series have suggested that anorexic patients are more likely to come from families of girls. This was not so in this sample. There were just as many families with more girls than boys than vice versa (41% in each case). There were no significant differences between the number of brothers and the number of sisters between the 3 groups. One AN family consisted of 8 children. "Family stage" is customarily defined as the median age of children in the family and can be a useful benchmark of the kind of lifestyle expected in that household. There was no significant difference between groups for family stage (Median 19, Range 11 to 32).

TABLE 6.2 FAMILY STRUCTURE

	Anorexic n = 27	Cystics n = 29	Well n = 31
Marital Status			
Married	85%	86%	94%
Widowed	4%	7%	0%
Divorced/Separated	11%	7%	7%
No of Children			
Mean (SD)	2.4(1.3)	2.3(0.7)	2.2(0.6)
Family Stage			
Median (Range)	19 (12-32)	20 (13-29)	17 (11-24)

6.1.5 Social Class

The social class breakdown of the families is shown in Table 6.3. Class was assessed by the Registrar General's classification of occupations (Office of Population Censuses and Surveys, 1980) using occupation of the head of household, which in most cases was the father. There were significant differences in social class between groups (Kruskal-Wallis ANOVA by ranks, $H = 8.23$, df 2, $p = .02$) with the cystic fibrosis families falling into lower socio-economic groups than the anorexic and well families. The skew towards the higher social classes found in most anorexia nervosa series (eg Russell et al, 1987; Gowers and McMahon, 1989) was found here with some 63% of families falling into classes I and II, 26% being the proportion expected for Scotland (General Register Office for Scotland, 1981). No AN families fell into classes IV and V. As 64% of the well families also fell into classes I and II, the planned social class match here was satisfactory. However, only 38% of the cystic fibrosis families fell into classes I and II.

TABLE 6.3 SOCIAL CLASS[1] OF FAMILIES

Class	Anorexics n = 27	Cystics n = 29	Well n = 31	Scotland[2]
I and II	63%	38%	64%	26%
IIIa	26%	21%	16%	11%
IIIb	11%	31%	13%	37%
IV and V	0%	13%	6%	25%

[1] Social Class of Head of Household (Registrar General's classification)
[2] General Register Office for Scotland 1981

6.2 Index Child Health

6.2.1 Index Child Health History

At the time of interview the AN patient had suffered from their illness for a median of 3 years ranging from 10 months to 8 years. By contrast the CF patient had been diagnosed for a median 17 years. 80% of CF patients have their diagnosis established by the age of 2 (Penketh, 1987) and this was the case in this series. This ranged from a patient who had been diagnosed only 2 years previous to one who had been diagnosed 24 years ago. Thus CF families on the whole had had many years to adapt to their child's illness whereas it had been a relatively recent problem for the AN families. The AN families all had a clear picture of "life before anorexia nervosa". The median number of admissions to hospital for treatment of the index illness was 1 (Range 0-5) in the anorexia sample and 4 (Range 0-50) in the cystic fibrosis sample.

44% of AN patients had a "mental health history" (other than AN) compared to 14% of CF patients and 7% of the well group (χ^2 = 13.3, p = .001). 41% of the AN group had received treatment at some time for a depressive illness compared to only 3% (1 case) in the CF sample.

6.2.2 Current Health Status of the Index Child

10 of the AN patients at the time of assessment no longer met the DSM-III-R weight criterion for Anorexia Nervosa which demands that they should weigh 85% or less of expected weight for height. 4 of the anorexic patients admitted to binge eating at least once a month. Of these 1 also met the DSM-III-R criteria for bulimia nervosa. 18 patients used excessive exercising as a means of losing weight on half or more days per month. 6 patients used either vomiting and/or laxative abuse at least once a month for the same reason. 3 AN patients had a concurrent diagnosis of major depressive episode (DSM-III-R) but none of the CF patients or well children met DSM-III-R criteria for depression.

The main findings are summarised in Table 6.4. Not surprisingly, the percentage of weight expected for height and age (%W/H) was significantly different across groups ($F = 12.8$, $p < .001$) with both anorexic and cystic fibrosis patients being thinner than the well children. AN patients were slightly thinner than the CF patients although the difference just failed to reach significance (Duncan's multiple range test AN v CF, $p = .08$).

TABLE 6.4 CURRENT HEALTH STATUS OF THE INDEX CHILD

	Anorexics n = 27	Cystics n = 29	Well n = 31
% Weight/Height[1,2] Mean (SD)	78.7 (15.1)	84.5 (12.0)	94.7 (9.2)
Body Mass Index Mean (SD)	17.4 (3.5)	18.9 (2.4)	20.9 (2.0)
Cooperman Scale Mean (SD)	-	5.3 (2.8)	-
FVC Mean (SD)	-	70.8 (6.0)	-
FEV_1 Mean (SD)	-	58.6 (6.0)	-
GHQ-30 Median Score[3,4]	8	3	1
Morgan and Russell Scales Median Scores[3]			
Food Intake	4	10	12
Menstrual[5]	0	12	12
Socio-Economic[6]	8	10	12
Psychosexual	7.5	10	10
Mental State	8	12	12
Average Outcome Score[1] Mean (SD)	6.0 (2.9)	10.1 (1.4)	10.9 (0.8)

[1] ANOVA Analysis of Variance. All p values < **.001**
[2] Post hoc comparison of means. Duncan's multiple range test
AN v CF p = .08
CF v Well p = **.002**
[3] Kruskal-Wallis ANOVA by ranks. All p values < **.001**
[4] Mann Whitney U test
AN v CF p = **.005**
CF v Well p = .34
[5] CF group n = 9
[6] Mann Whitney U test
CF v Well p = **.05**
AN v CF p = **.004**

6.2.3 Morgan and Russell Scales

(Table 6.4) The AN group had Morgan and Russell Average Outcome scores that were significantly lower than the CF and well groups ($F = 54.3$, df 2,84, $p < .0001$). As the distributions of the Morgan and Russell subscales were not normal, the non-parametric Kruskal-Wallis ANOVA for ranks test was used to compare differences between subscales and the Mann-Whitney U test was used for paired comparisons. Main effects for all the Morgan and

Russell subscales were highly significant ($p < .001$). An examination of the scales shows that in general the cystic patients and the well young people were indistinguishable with healthier levels of functioning than the AN patients. The socio-economic subscale measures social relationships, social activities and economic performance. Here the cystic patients functioned less well than the well subjects ($p = .05$) but in almost every case this was due to economic incapacity rather than to social difficulties. However, they still functioned better than the anorexic patients ($p = .004$). The other subscale where the CF patients functioned less well than the well young people was Food Intake. This was due to low weight rather than to disturbed eating behaviour.

6.2.4 General Health Questionnaire

(Table 6.4) The GHQ-30 is a measure of general psychological distress. What was noticeable was how psychologically healthy the CF sample were. The main effect for groups was highly significant (Kruskal- Wallis ANOVA for ranks, $H = 12.96$, $p = .002$). Paired comparisons showed that the anorexic patients had a higher level of distress than the well groups with median scores of 8 and 1 respectively. The difference between the CF group (score 3) and the well group was not significant. A score greater than 5 is customarily used as a cut off to indicate a high probability of a psychiatric diagnosis (Goldberg, 1986). Looking at the data in this way 58% of the anorexic patients fell into the "clinical" group compared with only 17% of cystic fibrosis patients and 19% of well children (Table 6.5). Mann et al (1983), using the GHQ28, found a rate of 19% probable psychiatric cases in a sample of London 13 year old schoolgirls. The findings cannot be explained by the unequal distribution of male and female subjects in the groups. Looking at girls alone, only 1/9 (11%) of CF girls compared to 11/25 (56%) of AN girls and 5/23 (22%) of well girls scored above 5.

TABLE 6.5 GENERAL HEALTH QUESTIONNAIRE GHQ30 PERCENTAGE SCORING ABOVE 5

	n	Anorexia Nervosa	Cystic Fibrosis	Well	Cox (1987)
Child	87	58%	17%	19%	30%
Father	74	30%	20%	35%	
Mother	86	48%	34%	17%	

6.2.5 Current Treatment

70% of the AN sample were "in treatment" at the time of interview. This included 2 patients who were attending their family doctor only. The rest were either outpatients or day patients. 19% of the AN sample had been in hospital in the 3 months prior to interview compared to 31% of the CF sample. 22% of the AN sample had attended at least one "family interview" in the previous 3 months compared to 4% (1 case) of the CF sample. A family interview was defined as a meeting with one or more parents present. While none of the CF sample was receiving psychotropic medication, 26% of the AN sample were taking anxiolytics or anti-depressants. 76% of the CF groups had taken antibiotics including 38% who had self administered I.V. antibiotics in the previous 3 months.

6.2.6 Physical Health Status of CF Patients

The health status of this group covered a wide range. The mean Cooperman score was 5.3 (SD 2.8). Lung function in this group was similar to the large Canadian study of psychological functioning of CF adolescents (Cowen et al, 1984). Mean Forced Vital Capacity (FVC) was 70.8 (SD 6.0) and Forced Expiratory Volume in one second (FEV_1) was 58.6 (SD 6.0). The healthiest boy in the study had a Cooperman score of 9, FVC of 141% and FEV_1 of 142%. His weight was 125% of what was expected for his age and height. He carried out twice daily physiotherapy, took pancreatic enzymes with every meal and had had a 3 week period of infection in the

previous 6 months when he had to miss school. Apart from this his daily functioning seemed unimpaired. He played rugby for his school and went hill walking most weekends. At the other extreme the illest patient was on the heart-lung transplant waiting list, had a Cooperman score of 1, FVC of 23% and FEV_1 of 15%. Her weight was 73% of what was expected. She had physio-therapy 3 times daily carried out by her mother as she was too weak to do this herself, was confined to her bedroom, could walk to the bathroom with effort, self administered I.V. antibiotics, spent most of her resting period using an oxygen cylinder and had only 2 hours per day on non health-related activities.

6.3 Parents

6.3.1 Psychiatric Status of the Parents

There was no significant difference in the proportion of mothers (18%) or fathers (9%) in the 3 groups who had a mental health history. "Mental health history" was defined as having received treatment from either family doctor or hospital specialist for psychological disorder or distress. As in the general population, more mothers than fathers had such a history.

On the GHQ-30, a measure of psychological distress, the fathers emerged as a healthy group with median scores in the AN group of 2 (Range 0-29); in the CF group of 1 (Range 0-20) and in the well group of 3 (Range 0-20). These small differences between groups did not reach significance (Kruskal-Wallis test ANOVA by ranks, H = 5.2, df 2, p = .07). The median scores for mothers were AN 5 (Range 0-20); CF 2 (Range 0-27) and well 1 (Range 0-15). These differences reached significance (Kruskal-Wallis H = 6.73, df 2, p = .03). Paired comparisons (Mann-Whitney U test) showed that the significant differences lay between the AN and well group only (p = .01). The difference between CF mothers and well mothers just failed to reach significance (p = .09). Looking at the same data categorically, 48% of the AN mothers had a high probability of psychiatric diagnosis compared to

34% of CF mothers and 17% of well mothers (Table 6.5). CF mothers had twice the rate of "caseness" compared to the well mothers but this difference would only reach significance if there was a sample size of at least 50 in each group. However, the well mothers were noticeably healthier than the 30% of Cox's British normal population study.

6.3.2 Impact on Family Scale (Stein and Riessman)

The results of this scale are summarised in Table 6.6. This scale measures the impact that chronic illness has on parents. Families were asked to rate the scale on the basis of their current experience. Families with partially recovered AN patients said that they would have given higher scores had they been asked to complete the scales at the height of the illness. Overall there were no significant differences for either mothers or fathers between the cystic fibrosis and the anorexic group. Looking at the individual subscales, both AN mothers and fathers felt that they had less mastery over the illness than did the CF parents (fathers $t = 2.76$, $p = .008$ and mothers $t = 2.3$, $p = .03$). This was in accord with the clinical impression given. CF mothers felt under more financial strain than the AN mothers ($t = 2.16$, $p = .04$) but this could be accounted for by the social class difference between groups. CF families have higher than average food bills and were often having to spend relatively large amounts on transport costs.

TABLE 6.6 IMPACT ON FAMILY SCALE

	Anorexia Nervosa Mean (SD)	Cystic Fibrosis Mean (SD)	t	p
Fathers	n = 22	n = 25		
Financial	10.0 (3.1)	11.1 (2.1)	1.50	.13
Social	15.9 (2.7)	16.8 (2.6)	1.30	.22
Personal Strain	15.2 (2.7)	15.8 (1.8)	0.95	.36
Mastery	8.7 (1.3)	7.6 (1.6)	−2.76	**.008**
Total	49.7 (6.5)	49.5 (6.6)	0.90	.37
Mothers	n = 26	n = 29		
Financial	9.8 (2.4)	11.5 (3.2)	2.16	**.04**
Social	16.3 (2.9)	17.5 (3.9)	1.2	.22
Personal Strain	16.3 (2.0)	17.3 (2.2)	1.7	.09
Mastery	8.8 (1.9)	7.7 (1.8)	−2.3	**.03**
Total	51.5 (6.8)	54.0 (7.9)	1.41	.18

6.4 Family Functioning - Self Report Measures

6.4.1 Family Assessment Measure

The Family Assessment Measure (FAM) is a self report measure that was completed by parents, index child and any siblings living at home aged 13 and over. The FAM was completed by all the index children and their mothers (n = 87), and by all but 2 fathers (n = 74) and by all but 2 siblings living at home (n = 44). Only 4 families had 3 or more children living at home aged 13 or over. Because of these small numbers, data was subject to individual analysis on one sibling only, in each case the eldest sibling being selected (Sibling 1). The General Score is the mean of the 7 FAM subscales. Table 6.7 shows that there were no significant differences in mean General Scale scores between the 3 groups for the index child, mothers, fathers or eldest sibling. The mean score of the cystic fibrosis siblings group appears to be high but the sample size was small (n = 14) and the standard deviation large.

TABLE 6.7 FAMILY ASSESSMENT MEASURE (GENERAL SCALE) MEAN SCORES

	Anorexia Nervosa Mean (SD)	Cystic Fibrosis Mean (SD)	Well Mean (SD)	F[1]	p
Index Child	52.1 (9.5)	51.3 (6.1)	50.5 (7.8)	0.29	.74
Mother	50.8 (6.0)	52.8 (4.7)	51.2 (6.5)	1.10	.33
Father	51.3 (4.9)	51.5 (3.8)	50.2 (5.7)	0.57	.57
Sibling 1	51.5 (7.5)	56.0 (13.3)	52.2 (6.9)	0.83	.44

[1] ANOVA Analysis of Variance.

Tables 6.8 to 6.11 show the FAM results for the subscales of the FAM. Multivariate analyses of variance (MANOVA) were carried out in preference to the univariate approach as the subscale scores intercorrelate. There was a small significant effect for the Index Child (Wilks' Lambda 0.70, df 18,152, p = .05). A post hoc comparison of means (Duncan's multiple range test) demonstrated that this effect was due to the Social Desirability subscale only in which the anorexia nervosa children were less likely to give socially desirable answers than the cystic fibrosis group (Table 6.8). The Denial and Social Desirability subscales do not form part of the General Scale.

TABLE 6.8 FAMILY ASSESSMENT MEASURE (INDEX CHILD) INDIVIDUAL SUBSCALES

	Anorexia Nervosa n = 27 Mean (SD)	Cystic Fibrosis n = 29 Mean (SD)	Well n = 31 Mean (SD)
Task Accomplishment	53.1 (13.5)	50.0 (9.0)	50.3 (10.4)
Role Performance	50.5 (11.3)	49.5 (7.0)	51.6 (8.1)
Communication	52.7 (10.8)	53.1 (7.7)	48.7 (9.0)
Affective Expression	53.3 (10.2)	53.2 (8.8)	53.6 (8.3)
Affective Involvement	53.0 (13.0)	53.0 (8.5)	50.7 (10.9)
Control	50.5 (10.3)	50.6 (6.2)	49.7 (8.1)
Value and Norms	51.8 (9.1)	49.6 (8.1)	49.3 (9.4)
Social Desirability[1]	44.8 (8.3)	51.2 (7.6)	48.7 (9.9)
Denial	43.0 (10.0)	45.6 (8.3)	46.6 (9.6)

MANOVA. Wilks' Lambda 0.70, df 18,152, p = **.05**
[1] Post-hoc Comparison of Means. Duncan's Multiple Range Test
AN v CF p = **.04**

A significant effect was found between mothers (Wilks' Lambda = 0.62, df 18,152, p = .004) for the FAM subscale. Post hoc comparison of means again showed that the anorexia nervosa mothers were less likely to give socially desirable responses than the cystic fibrosis mothers (p = .003). (Table 6.9). For fathers and sibling 1, main effects for subscales were all non-significant (Tables 6.10 and 6.11).

TABLE 6.9 FAMILY ASSESSMENT MEASURE (MOTHER) INDIVIDUAL SUBSCALES

	Anorexia Nervosa n = 27 Mean (SD)	Cystic Fibrosis n = 29 Mean (SD)	Well n = 31 Mean (SD)
Task Accomplishment	49.5 (8.8)	49.2 (5.6)	50.7 (9.7)
Role Performance	52.1 (9.5)	55.3 (9.5)	53.6 (8.5)
Communication	49.4 (7.0)	52.7 (5.6)	50.5 (8.4)
Affective Expression	51.3 (9.7)	54.1 (7.9)	49.5 (6.9)
Affective Involvement	51.1 (6.5)	52.7 (6.6)	49.9 (7.7)
Control	50.5 (8.7)	53.4 (5.8)	50.5 (9.2)
Value and Norms	51.4 (7.1)	52.4 (8.2)	52.6 (7.1)
Social Desirability[1]	47.4 (5.1)	52.7 (6.8)	50.3 (7.2)
Denial	49.8 (9.1)	49.6 (7.0)	46.1 (5.9)

MANOVA. Wilks' Lambda = 0.62, df 18,152, p = **.004**
Post-hoc Comparison of Means. Duncan's Multiple Range Test
[1] AN v CF p = **.02**
AN v Well p = .09

TABLE 6.10 FAMILY ASSESSMENT MEASURE (FATHER) INDIVIDUAL SUBSCALES

	Anorexia Nervosa n = 23 Mean (SD)	Cystic Fibrosis n = 25 Mean (SD)	Well n = 26 Mean (SD)
Task Accomplishment	47.6 (8.1)	49.4 (8.1)	47.8 (7.7)
Role Performance	53.1 (7.2)	50.1 (10.5)	50.3 (7.1)
Communication	51.5 (8.0)	52.3 (5.0)	51.1 (6.9)
Affective Expression	53.7 (7.1)	54.3 (4.7)	52.0 (6.0)
Affective Involvement	52.5 (6.8)	51.9 (6.6)	48.3 (9.0)
Control	49.7 (7.7)	51.6 (5.6)	51.0 (8.0)
Value and Norms	51.2 (8.9)	50.8 (6.6)	50.3 (8.0)
Social Desirability	48.1 (5.9)	52.4 (6.7)	51.7 (7.7)
Denial	51.4 (10.3)	49.7 (8.3)	47.7 (8.8)

MANOVA. Wilks' Lambda = 0.67, df 18,126, p = .08

TABLE 6.11 FAMILY ASSESSMENT MEASURE (SIBLING 1) INDIVIDUAL SUBSCALES

	Anorexia Nervosa n = 13 Mean (SD)	Cystic Fibrosis n = 14 Mean (SD)	Well n = 13 Mean (SD)
Task Accomplishment	50.1 (7.5)	53.8 (8.7)	51.2 (9.6)
Role Performance	52.3 (5.1)	51.6 (3.5)	54.4 (7.0)
Communication	51.4 (13.6)	52.9 (6.7)	55.9 (10.1)
Affective Expression	52.2 (7.6)	53.2 (11.0)	51.5 (8.4)
Affective Involvement	51.8 (10.0)	54.7 (11.6)	50.3 (9.9)
Control	51.3 (8.8)	51.4 (6.1)	52.3 (7.0)
Value and Norms	51.1 (9.4)	51.2 (4.1)	50.3 (7.4)
Social Desirability	46.9 (8.9)	49.5 (7.5)	46.5 (11.5)
Denial	49.8 (9.7)	45.9 (10.5)	42.5 (7.7)

MANOVA. Wilks' Lambda = 0.55, df 18,156, p = .43

Table 6.12 shows the correlations between the FAM scores of family members. If individuals within the family are describing the same phenomenon, then there should be some relationship between family scores. The strongest relationship is between parents ($r = .52$, $p < .001$) followed by mothers and index child ($r = .33$, $p = .002$) and fathers and index child ($r = .26$, $p = .02$). There was no agreement, however, between siblings and the index child but sample size was small.

TABLE 6.12 FAMILY ASSESSMENT MEASURE (GENERAL SCALE) CORRELATIONS BETWEEN FAMILY MEMBERS

	n	Index Child	Mother	Father
Index Child	87	-		
Mother	87	.33**	-	
Father	75	.26*	.52***	-
Sibling 1	40	.08	.31*	-.05

* p < **0.05**; ** p < **0.01**; *** p < **0.001**
Pairwise deletion of missing data

By averaging the FAM scores for each family, one can arrive at an estimate of the overall family atmosphere, the household score (Table 6.13). The mean score of the whole sample was 51.75 (SD 4.98) with no differences between groups. Skinner (1983) suggested a score of 60 (more than one standard deviation) should be the cut off point for family dysfunction. Only 4 cases scored 60 or more. When one looked at the families who scored 56 or more (one standard deviation above the mean of this sample), 22% of the AN families, 16% of the well families and 10% of the CF families could be described as high scorers. The high scoring families were clinically recognisable. They tended to be oppositional, opinionated families with a high level of open quarrelling.

TABLE 6.13 FAMILY ASSESSMENT MEASURE: HOUSEHOLD

n = 87		
AN Mean (SD)	CF Mean (SD)	Well Mean (SD)
51.7 (5.1)	52.7 (4.1)	50.9 (5.3)

ANOVA. Analysis of Variance, df 2,84, F = 0.95, p = .39

How did the FAM Scale results compare with the Scottish norms established in the Schools Project (Chapter 9) ? The scores of the 214 adults (Mean 50.4, SD 6.3) and 214 young people (Mean 51.3, SD 7.5) who took part in that study differ little from the mean scores of the well families in this sample. Looking at the 2 subscales where some differences between groups were found, Social Desirability and Affective Expression, the Scottish norms again mirror very closely the norms of the well sample which gives support to the notion that the well families were a representative sample.

A correlation of .45 ($n = 87$, $p < .0001$) was found between the level of emotion distress (GHQ) in the index child and that child's FAM general score. A weaker relationship ($r = .22$, $p = .05$) was found between mother's GHQ and her FAM general score but there was no relationship for fathers.

6.4.2 The Edinburgh Family Scale

Table 6.14 shows the mean scores for the 3 "psychosomatic" family dimensions: conflict, enmeshment and rigidity for the index child. Multivariate analysis of variance showed an overall effect (Wilks' Lambda = 0.79, df 6,164, $p = .002$). Post hoc comparison of means (Duncan's multiple range test) showed that the AN patients were more likely to describe themselves as enmeshed than were the CF patients ($p = .05$).

Girls were more likely to describe themselves as enmeshed than boys ($r = .31$, $p = .004$) and young people from lower social classes were more likely to describe themselves as enmeshed ($r = .22$, $p = .04$). The samples (Section 6.1.1 and 6.1.5) were not adequately matched on these variables. However, the relationship between AN and enmeshment still held when these 2 variables were held constant. The CF young people described themselves as having more rigid families than AN ($p = .06$) and well families ($p = .03$). There was no difference in the 3 conflict scores.

TABLE 6.14 EDINBURGH FAMILY SCALE: INDEX CHILD

	AN n = 26 Mean (SD)	CF n = 29 Mean (SD)	Well n = 31 Mean (SD)
Conflict	52.5 (13.4)	48.7 (9.1)	50.0 (11.1)
Enmeshment[1]	55.6 (9.4)	48.9 (9.5)	51.9 (9.5)
Rigidity[2]	48.1 (9.8)	53.8 (6.4)	49.1 (8.4)

MANOVA Wilks' Lambda = 0.79, df 6,162, p = **.002**
Post-hoc comparison of means. Duncan's Multiple Range Test
[1] AN v CF p = **.05**
AN v Well p = .15
[2] CF v Well p = **.03**
CF v AN p = .06

Tables 6.15 and 6.16 show that for mothers and fathers, the means for all 3 subscales were not significantly different. Table 6.17 shows a significant overall effect for the elder sibling (Wilks' lambda = 0.62, p = .03) but univariate analysis of the subscales with post hoc comparison of means failed to confirm this. The mean scores for all eligible family members, the household score, is shown in Table 6.18. There were no significant differences between households.

TABLE 6.15 EDINBURGH FAMILY SCALE: MOTHER

	Anorexia Nervosa n = 27 Mean (SD)	Cystic Fibrosis n = 29 Mean (SD)	Well n = 31 Mean (SD)
Conflict	52.1 (9.8)	52.7 (7.6)	53.0 (11.1)
Enmeshment	51.0 (11.5)	53.8 (9.1)	51.1 (11.6)
Rigidity	45.6 (9.6)	50.0 (8.2)	46.8 (7.2)

MANOVA. Wilks' Lambda = 0.94; df 6,164, p = .53

TABLE 6.16 EDINBURGH FAMILY SCALE: FATHER

	Anorexia Nervosa n = 23 Mean (SD)	Cystic Fibrosis n = 25 Mean (SD)	Well n = 27 Mean (SD)
Conflict	51.3 (9.2)	51.9 (8.6)	49.9 (11.0)
Enmeshment	50.0 (9.9)	50.4 (11.0)	51.9 (10.8)
Rigidity	47.4 (9.1)	51.4 (7.7)	52.0 (9.1)

MANOVA. Wilks' Lambda = 0.93, df 6,140, p = .57

TABLE 6.17 EDINBURGH FAMILY SCALE: SIBLING 1

	Anorexia Nervosa n = 13 Mean (SD)	Cystic Fibrosis n = 12 Mean (SD)	Well n = 9 Mean (SD)
Conflict	47.6 (10.3)	52.3 (10.2)	53.1 (8.1)
Enmeshment	48.9 (11.2)	55.8 (12.0)	51.7 (7.8)
Rigidity	36.2 (7.1)	43.8 (9.6)	41.0 (8.0)

MANOVA. Wilks' Lambda = 0.62, df 6,58, p = **.03**
ANOVA. Conflict F = 1.06 p = .35
Enmeshment F = 2.65 p = .09
Rigidity F = 1.30 p = .28

TABLE 6.18 EDINBURGH FAMILY SCALE: HOUSEHOLD

	Anorexia Nervosa n = 27 Mean (SD)	Cystic Fibrosis n = 29 Mean (SD)	Well n = 31 Mean (SD)
Conflict	52.0 (7.8)	51.7 (6.8)	51.4 (8.3)
Enmeshment	51.9 (7.5)	51.4 (7.2)	52.0 (7.2)
Rigidity	46.0 (6.2)	50.8 (4.8)	48.6 (6.1)

MANOVA Wilks' Lambda = 0.88, df 6,164, p = .10

6.5 FAMILY FUNCTIONING: EXPRESSED EMOTION

Turning from the self report measures to observational measures, Tables 6.19 and 6.20 show the Expressed Emotion scores for mothers and fathers. As the distribution of scores is so skewed, results were analyzed as medians rather than means. However, in order to facilitate comparison with other published Expressed Emotion and eating disorder studies, the means and standard deviation EE are presented in Table 6.21.

6.5.1 Critical Comments

In all 3 groups both mothers and fathers made a median number of one critical comment only. Median scores for hostility were zero and indeed only 3 hostile remarks were made at all.

TABLE 6.19 EXPRESSED EMOTION: MOTHER

	Anorexia Nervosa n = 26 Med(Range)	Cystic Fibrosis n = 29 Med(Range)	Well n = 31 Med(Range)	H[1]	P
Critical Comments	1 (0-5)	1 (0-10)	1 (0-6)	0.92	.63
Hostility	0 (0)	0 (0-1)	0 (0)	-	-
EOI[2]	2 (0-4)	2 (0-5)	0 (0-2)	25.97	**<.001**
Positive Remarks	1 (0-5)	1 (0-6)	1 (0-7)	1.31	.51
Warmth	2 (0-4)	2 (0-5)	3 (0-4)	1.58	.45

[1] Kruskal-Wallis. ANOVA by Ranks.
[2] Mann Whitney U test AN v CF N.S.
CF v Well $p < .001$
AN v Well $p < .001$

TABLE 6.20 EXPRESSED EMOTION: FATHER

	Anorexia Nervosa n = 21 Med(Range)	Cystic Fibrosis n =22 Med(Range)	Well n = 26 Med(Range)	H[1]	P
Critical Comments	1 (0-4)	1 (0-8)	1 (0-3)	1.48	.48
Hostility	0 (0-3)	0 (0-1)	0 (0)	1.23	.54
EOI	0 (0-4)	1 (0-4)	0 (0-1)	5.99	.07
Positive Remarks	1 (0-5)	0 (0-6)	1 (0-6)	1.37	.50
Warmth	2 (0-4)	2 (0-4)	2.5(0-5)	1.25	.54

[1] Kruskal-Wallis. ANOVA by Ranks.

Compared with the schizophrenia studies, the level of criticism in these families is very low. Using the traditional cut-off of 6 or more critical comments to classify a parent as "high EE", only 3 mothers and 1 father in the whole sample could be classified as "high EE" and none of these were AN parents. Other studies of eating disorders (van Furth, 1991; le Grange, 1992a, 1992b) have given a high EE classification to cases where 2 or more critical comments were made. 36% of all mothers and 33% of all fathers could be classified as "high EE" on this basis.

TABLE 6.21 EXPRESSED EMOTION MEAN SCORES

	Anorexia Nervosa Mean (SD)	Cystic Fibrosis Mean (SD)	Well Mean (SD)
Mother	n = 26	n = 29	n = 31
Critical Comments	1.62 (1.5)	1.72 (2.4)	1.29 (1.3)
EOI	2.04 (1.2)	1.93 (0.4)	0.51 (0.7)
Father	n = 21	n = 22	n = 26
Critical Comments	1.29 (1.1)	1.1 (1.8)	1.04 (1.1)
EOI	1.10 (1.3)	0.77 (1.0)	0.27 (0.5)
% of high EE households[1]	42%	34%	3%

[1] $\chi^2 = 13.7$, df 2, p = **.001**

6.5.2 Emotional Overinvolvement

The mothers of both the illness groups showed significantly more overinvolvement than did the well group mothers (p = .001). Median EOI scores for fathers were very low and did not show a significant effect. High emotional overinvolvement has customarily been defined as a score of 3 or more. Analysing the data in this way, none of the well parents could be described as high EOI compared to 35% and 31% of the AN and CF mothers respectively. ($\chi^2 = 12.94$, df 2, p .002). 19% of AN fathers were high EOI compared to 4.5% of CF fathers and none of the well fathers. ($\chi^2 = 6.62$, df 2 p =.04).

No relationship was found between the number of critical comments made and emotional overinvolvement (Spearman's r = –.06, NS). A correlation of .37 (p = .002) was found between emotional over-involvement in mothers and fathers.

A correlation of .42 (n = 85, p = .0001) was found between maternal GHQ

and maternal EOI and of .29 (n = 68, p =.02) between paternal GHQ and paternal EOI. This relationship still held when weight (a rough measure of illness severity: see section 6.5.5) was held constant (mothers, r = .32, p = .003; fathers, r = .27, p = .03).

6.5.3 EE Household

A high EE household was defined in the traditional way (Leff and Vaughn, 1985) as one in which either parent obtains an EOI score of 3 or more and/or either parent obtains a high (6 or more) critical comments score. Using this definition 42% of AN households, 34% of CF households and 3% (1 case) of well households could be described as high EE (χ^2 = 13.17, df 2, p = .001). (Table 6.21)

6.5.4 Warmth and Positive Remarks

There were no significant differences in the amount of warmth or the number of positive remarks made between groups (Tables 6.19 and 6.20).

6.5.5 Expressed Emotion and Severity of Illness

There are intrinsic difficulties in comparing severity of illness in CF and AN patients as severity is best evaluated with different constellations of indicators in the 2 illnesses. There is however one indicator that the 2 illnesses have in common: weight. In CF patients there was a correlation (r = .71, p < .0001) between weight and the Cooperman Scale and between weight and FEV_1 (r = .81, p < .0001). In AN patients there was correlation (r = .69, p < .0001) between weight and the Morgan and Russell Average Outcome score (AOS). Therefore, although low weight may have a different meaning for AN and CF families, the fact that it correlates well with other indices of illness severity for the 2 illnesses and that in this series there were no significant differences between AN and CF patients for weight,

suggests that weight is not a meaningless operationalisation of illness severity.

There was a significant relationship between emotional overinvolvement and weight (n = 55, Spearman's r = –.38, p = .004) in the 2 ill groups but no association in the well group (n = 31, r = –.06, NS)

In the CF group a composite variable "CF illness severity" was derived by calculating the mean of weight and the 2 lung function variables, FVC and FEV_1. All 3 variables are expressed as a percentage of expected score for age and height. The greater loading that this composite variable gives to lung function is in accordance with the clinical importance of lung function compared to weight as a prognostic sign. Using this index of health, a significant relationship was found once again between illness severity and EOI (n = 29, Spearman's r = –.37, p = .05) in CF households. In AN households there was a significant relationship between the Morgan and Russell Average Outcome Scores and EOI (n = 27, Spearman's r = .62, $p < .001$).

All these findings suggest that EOI may be an illness related factor and offer an explanation as to why high EOI was more common in the ill groups than in the well group.

By contrast no relationship was found between the number of critical comments made either by parent or by combined household and severity of illness measured by the 3 methods above.

6.6 Family Functioning - Problem Solving Task

3 concepts were measured using the audiotaped interaction of family triads: "overprotection", "generation boundaries" and problem solving skills. The median "overprotection" scores are shown in Table 6.22. There was a significant main effect between groups (Kruskal-Wallis H = 6.45, p = .04).

Paired comparisons (Mann-Whitney U test) showed that while there was no difference between the CF and the well families, the AN families showed significantly more overprotective behaviour than the other groups. Overprotection scores could range from 0 to a maximum of 20. A score of 6 or more indicates that at least 3 clear examples of overprotective/enmeshed behaviour have been observed. Looking at the same data categorically, score 6 or more was observed in 44% of AN families compared to 19% and 10% of CF and well families respectively.

Generation boundaries score could range from 0 - 4 with 0 to 1 suggesting no evidence of weak boundaries and scores of 2+ suggesting some evidence. Not many families show evidence of weak generational boundaries, median scores for all 3 groups being zero. However, even with this low level, the CF families show significantly less evidence of weak boundaries than the well or AN groups (Kruskal-Wallis H = 8.30, p = .02). Only 7% of CF families compared to 24% of AN families and 33% of well families showed some evidence of weak boundaries (Table 6.22).

TABLE 6.22 PROBLEM SOLVING TASK OVERPROTECTION AND BOUNDARIES

	Anorexia Nervosa n = 25 Med(Range)	Cystic Fibrosis n = 27 Med(Range)	Well n = 30 Med(Range)	H[1]	P value
Overpro-tection[1]	5 (0-17)	1 (0-16)	1.5(0-8)	6.45	**.04**
Bound-aries[2]	0 (0-4)	0 (0-3)	0 (0-4)	8.30	**.02**

[1] Kruskal-Wallis Test ANOVA by Ranks
[2] Mann-Whitney U Test AN v CF p = **.04**
AN v Well p = **.02**
[3] Mann Whitney U Test CF v Well p = **.03**
CF v AN p = **.03**

Table 6.23 shows the problem solving ability of the 3 groups. With families classified into good, medium and poor problem solvers, the AN families were

slightly less effective than the other groups (χ^2 = 10.2, df 4, p = .04). However, this was not due to greater use of conflict diffusion or avoidance. There were families who actively avoided conflict but they were divided equally between groups (Table 6.24).

TABLE 6.23 PROBLEM SOLVING TASK - PROBLEM SOLVING SKILLS

	Anorexia Nervosa n = 27	Cystic Fibrosis n = 25	Well n = 31
Good	12%	48%	29%
Medium	32%	19%	39%
Poor	56%	33%	32%

χ^2 = 10.2, df 4, p = **.04**

TABLE 6.24 PROBLEM SOLVING TASK MINUCHIN'S CONFLICT AVOIDANCE

	Anorexia Nervosa n = 27	Cystic Fibrosis n = 25	Well n = 31
No avoidance	68%	81%	74%
Some avoidance	20%	7%	16%
Strong avoidance	12%	11%	10%

χ^2 = 1.9, df 4, p = .75

6.7 Life Events

Table 6.25 shows the incidence of life events in the previous 6 months. The pattern of events is similar to that found in the schools study (Chapter 9) with important exams and trouble with relatives being the most common events. There were no significant differences in the frequency of families who had experienced one or more severely threatening events in the previous 6 months. The incidence for the CF families was 14%, AN families 13% and well families 28%. Interestingly the finding that this group of well

families tended to have more severe events is in the opposite direction from that expected from most life event research. This rate of threatening events is very similar to the 15% Brown and Harris (1989) found in a community survey.

The experience of several negative life events might be expected to have some impact on family functioning. However, there was no correlation between the presence or absence of a severely threatening event and FAM scores, EFS scale scores or Expressed Emotion scores.

TABLE 6.25 LIFE EVENTS IN PAST SIX MONTHS (n = 87)

Has this happened to anyone in your family circle in the past six months?	Incidence of event	Incidence of severe threat
1. Retired from work	9	1
2. Started or left a job	23	2
3. Started or left school or college	17	0
4. Had trouble at work	13	4
5. Had a major change in work conditions or responsibilities	19	2
6. Sat important exams	41	2
7. Got engaged or married	7	1
8. Family member left home	10	1
9. Became pregnant	2	0
10. Had a baby	6	0
11. New person joined the household	4	1
12. Had trouble with relatives (outside household)	10	0
13. Death of person in household	0	0
14. Death of other close family member or friend	13	3
15. Major accident or injury	1	0
16. Major change in health of family member (other than child in research study)	12	2
17. Major change in quality of marital relationship	3	1
18. Divorce or break up of special relationship	4	2
19. Major change in financial situation	14	3
20. Loss, damage or theft of property	12	0
21. Had trouble with the police or law	0	0
22. Moved house or change in housing conditions	8	2

6.8 Relationships between the Household Climate Variables

A number of different variables were used to measure the emotional climate of the household and a correlation matrix of these variables is found in Table 6.26. Mean family scores for all members of the household aged 13 and over were calculated for the Enmeshment, Conflict and Rigidity subscales of the Edinburgh Family Scale (EFS) (MESH, CON AND RIG) and the Family Assessment Measure (FAM). If either parent obtained a score of 2 or more critical comments on the Expressed Emotion Index, the household was defined as high critical comments (HI-CC). If either parent had an EE score of 3 or more, the household was defined as high emotional overinvolvement (HI.EOI). Finally Overprotection (OVER) and poor problem solving skills (PROB) measured from the problem solving task were included.

Of interest in the matrix is the high correlation between the FAM scale and the conflict scale ($r = .81$, $p < .001$) suggesting that they are measuring much the same thing. High levels of self reported enmeshment correlated negatively with high levels of conflict ($r = -.51$, $p < .001$). There was a small positive correlation between rigidity and enmeshment ($r = .25$, $p = .02$). In other words families who see themselves as having no difficulty in resolving conflict also see themselves as close and cohesive and disliking change. These findings mirror exactly the findings in the school population on which the Edinburgh Family Scale was developed and the FAM validated (see Chapter 9, School Project).

TABLE 6.26 EMOTIONAL CLIMATE OF THE HOUSEHOLD CORRELATIONS[1] BETWEEN VARIABLES

n = 87 (Missing data pairwise deleted)

	MESH	CON	RIG	FAM	HIEOI	CC	OVER
MESH	1						
CON	**-.51**	1					
RIG	**.25**	-.12	1				
FAM	**-.53**	**.81**	-.12	1			
HIEOI	**.29**	-.04	-.09	-.03	1		
CC	-.11	.19	-.13	**.24**	-.05	1	
OVER	.02	.00	-.14	.02	**.39**	-.06	1
PROB	-.02	**.24**	-.19	.17	.14	**.24**	.07

[1] Spearman's r
All figures in bold $p < .02$

Key

MESH	-	Edinburgh Family Scale Enmeshment Mean Family Score
CON	-	Edinburgh Family Scale Conflict Mean Family Score
RIG	-	Edinburgh Family Scale Rigidity Mean Family Score
FAM	-	Family Assessment Measure Mean Family Score
HIEOI	-	Expressed Emotion Overinvolvement
HICC	-	Expressed Emotion Critical Comments
OVER	-	Problem Solving Task - Overprotection
PROB	-	Problem Solving Task Poor Problem Solving Skills

In order to gain a more economical understanding of the relationships between these household climate variables, a principal components analysis was performed. Rigidity was excluded due to low intercorrelations with the other variables. After Varimax rotation, 3 factors emerged with eigenvalues greater than 1 (Table 6.27).

TABLE 6.27 PRINCIPAL COMPONENTS ANALYSIS OF HOUSEHOLD CLIMATE VARIABLES AFTER VARIMAX ROTATION

(n = 87)

	Factor I[1]	Factor II	Factor III
Variance	35%	21%	15%
MESH[2]	**-.79**	-.18	-.09
CON	**.89**	.02	-.17
FAM	**.90**	-.03	-.14
HIEOI	-.14	**-.85**	-.06
OVER	.10	**-.80**	.01
HICC	.16	.11	**-.75**
PROB	.10	-.16	**-.78**

[1] Factor loadings > .30 in bold
[2] See Table 6.26 for key to variables

The first factor consists largely of negative self evaluation: high levels of EFS conflict and FAM, low levels of EFS enmeshment. The second factor taps observed levels of overprotection/ overinvolvement derived from the Expressed Emotion EOI score and the Problem Solving Task overprotection score. The third factor loads on observed criticism and poor problem solving ability derived from Expressed Emotion Critical Comments and from observations of poor problem solving ability. These 3 factors together explain 71% of the variance.

The factor loadings were used to derive 3 new variables. Differences between the 3 groups on these household climate factors are shown in Table 6.28. Using the Median test with χ^2, Factor II is the only variable to show significant differences, both ill groups showing considerably more overprotection/overinvolvement than the well group ($\chi^2 = 16.43$, $p = .0003$). The difference between proportions in the AN and CF groups did not reach significance.

TABLE 6.28 EMOTIONAL CLIMATE - 3 FACTORS MEDIAN TEST[1]

	AN n = 27	CF n = 29	Well n = 31	χ^2	p
Factor I[2]	52%	48%	48%	.09	.95
Factor II	74%	59%	23%	16.43	**.0003**
Factor III	59%	45%	45%	2.4	.30

1 % Scores > Median

2 Factor I negative self evaluation of family
Factor II observed overprotection/enmeshment
Factor III observed poor problem solving skills and criticism

6.9 Predictors of Household Climate

What were the risk factors for dysfunctional household climate in the homes of sick children? A number of variables are prima facie potentially important predictors of poor climate. The severity of the illness, the perceived burden of illness, the emotional distress of individual family members are likely contenders (see Chapters 2 and 3 for literature reviews).

Perceived burden of illness was operationalised by the maternal and paternal Impact on Family scores, emotional distress by the GHQ scores of index child, mother and father, and household climate by the 3 derived factors. Severity of illness was operationalised by percentage of weight expected for height and age (% W/H).

The correlation between these variables is shown in Table 6.29.

TABLE 6.29 CORRELATIONS BETWEEN HOUSEHOLD CLIMATE, BURDEN OF ILLNESS, SEVERITY OF ILLNESS AND EMOTIONAL DISTRESS

n = 56 (missing data pairwise deleted)

	Factor I	Factor II	Factor III
% W/H	.08	**.40**	.12
Impact on Family (F)	**.32**	-.04	-.05
Impact on Family (M)	.14	-.17	.08
GHQ Father	.05	-.06	-.06
GHQ Mother	**.32**	**-.29**	-.03
GHQ Index Child	.14	-.23	.06

All figures in bold $p < .03$.

The table shows that none of the variables had any relationship with observed criticism and poor problem solving skills (Factor III). Negative self evaluation of family (Factor I) correlated with high levels of paternal evaluation of burden of illness ($r = .32$; $p = .03$) and with maternal GHQ ($r = .32$, $p = .02$). Factor II correlated with weight ($r = .40$; $p = .002$) and with maternal GHQ ($r = -.29$, $p = .03$) In other words, the more enmeshment/overinvolvement was observed in the family, the more likely was the child to be of low weight. This applied to both AN and CF families. A multiple regression of Factor I on paternal Impact of illness and maternal GHQ entered together found that only 19% of the variance was accounted for.($F = 5.14$, df 2,44, $p = .01$, $R^2 = .19$). The regression of Factor II on weight, maternal GHQ and index child's GHQ entered together found that 20% of the variance was accounted for by weight alone. ($F = 3.61$, df 3,43, $p = .03$, $R^2 = .20$)

Not too much should be made of these correlations. Enthusiasm for the highest correlation, that between weight and Factor II ($r = .40$) wanes somewhat when it is noted that the 95% confidence interval ranges from $r = .15$ to $r = .54$.

CHAPTER 7: THE FOLLOW-UP STUDY

7.1 Procedure

It was planned for the sample to be followed up 8 months after the baseline interviews had taken place. Contact was made by telephone with all families about a month before the 8 months (35 weeks) target date. With the ill sample families, contact was first made with the index child's therapist/physician.

Because the initial interviews had not been completed by the time the follow up phase began, the volume of work was too great to permit the well families to be reinterviewed. Travel to a family's home and carrying out the interviews was at least half a day's work. Again due to the volume of work, the independent assessor[1] was not able to visit the index child again so that the Morgan and Russell scales were completed by the author (See Section 7.6.1 for bias checks). It was not necessary on this occasion to elicit information from the index child for the Problem Solving Task which had been one of the main tasks of the independent assessor.

All families (well sample included) had repeat questionnaires posted out which were either posted back or collected at interview. If well families had not returned their questionnaires within 3 weeks, they were reminded by letter. If this produced no response, they were contacted by telephone.

Data on current lung function was collected from the CF clinics.

One interview only was carried out with each of the ill group families. At the interview, the Camberwell Family Interview and the Problem Solving Task were repeated. There were modifications to both. The follow-up CFI was in practice slightly shorter than the initial CFI. Families were asked to report on events since the last interview but were not asked to repeat in

[1] Mrs Wilma Warwick

depth the earlier history of the illness. There was a greater opportunity for families to discuss any child rearing difficulties. However, the format of the interview was unchanged.

In the problem solving task, instead of problems suitable for discussion being elicited from the families, triads were asked to have another attempt at discussing the first problem tackled at the baseline assessment. For the second problem, triads were asked to "plan a menu together" following the instructions exactly of Minuchin's family task (Minuchin, 1978, p 335). Minuchin felt that "planning a menu" was the most important medium for eliciting his "psychosomatic family" dimensions. This task has the disadvantage that the AN families become immediately identifiable from the tape so that ratings cannot be made blind to group. However, this disadvantage has to be traded off against the possibility that the "psychosomatic family" might have been missed at the baseline interview.

7.2 Attrition

As in all follow-up studies, there was some loss of cases and some loss of data. Case loss (no data at all) was 6/87 subjects (7% of the sample). 1 CF patient had died and a second whom I had been asked not to recontact by the physician died a month after her follow-up date. 2 CF families cancelled several follow-up appointments before finally refusing. The 1 missing AN family had refused to be interviewed at baseline but had completed questionnaires. In the well group, 1 index child and her mother had done a moonlight flit from home abandoning the father. Through some detective work, we did manage to trace them but they were vexed to have been "discovered" and refused to take part.

Thereafter success in data collection varied according to the methodology. Missing data at follow-up (including the 6 cases described above) is summarised in Table 7.1. It can be seen from the table that questionnaire completion was more fruitful than was the problem solving task which

involved gathering together the family triad of mother, father and index child.

TABLE 7.1 PERCENTAGE OF BASELINE SAMPLE WITH MISSING DATA AT FOLLOW-UP

	CF $n^1 = 29$	AN n = 27	Well n = 31
Questionnaires	16%	4%	6%
EE	14%	19%	N/A
Problem Solving	28%	26%	N/A

n^1 Baseline sample

27% of the ill sample did not take part in the problem solving task at follow-up. In order to check for bias in the drop-out group, an attempt was made to fit a logistic regression model (logit) that would predict drop-outs from baseline data. The 3 family functioning factors, negative self evaluation, observed enmeshment and overinvolvement, and observed criticism and poor problem solving skills did not predict drop-out for either AN or CF patients. Severity of illness assessed by Morgan and Russell scales did not predict drop-out in the AN group. However, in the CF group, severity of illness measured by the composite weight and lung function index was a predictor of drop-out. The odds ratio was 1.08. To make these ideas more concrete, for example, a boy whose weight and lung function were only 45% of what was expected for age and height had a 62% chance of not taking part in the follow-up problem solving task whereas a boy whose weight and lung function was 65% of what was expected had a 20% chance of dropping out.

The most common reason for not taking part in follow-up problem solving was teenage contrariness. I would arrive at the house by appointment to find the young person on the point of leaving the house for a party, snooker match etc. This was equally likely to happen in CF or AN households.

7.3 Date of Interview

The target follow-up date was 8 months (35 weeks). For the well group this was operationalised as the date when questionnaires were returned. There were significant differences between groups in follow-up date ($F = 10.06$, $p < .001$) with the CF sample being interviewed at 34 weeks (SD 4 weeks), the AN sample at 32 weeks (SD 3 weeks) and the well sample returned questionnaires at 37 weeks (SD 5 weeks).

7.4 Structure of Household

In several households, the household structure had changed. For example, teenagers had left home and others had returned and grandmothers had moved in or died. In order to measure household climate, the same criteria of "household" as at baseline was used for family members other than the index child although the individual members contributing to that climate may have changed. The index child was asked what percentage of time he/she had spent under the parental roof in the previous 3 months. 76% of the CF sample, 69% of the AN sample had spent every night at home compared to only 45% of the well sample. 22% of the total sample no longer met the original "living at home" criterion which was to have spent at least a third of their nights with their parents.

7.5 Treatment in the Previous Three Months

The treatment data is summarised in Table 7.2.

7.5.1 Anorexia Nervosa

27% of the AN sample (7 cases) had had no contact with any treatment agencies on AN related issues in the 3 months prior to follow-up interview. All these cases were at the "well" end of the AN spectrum. 23% of the sample (6 cases) had had either in-patient or day patient hospital contact

which ranged from a patient who attended hospital 2 days a week to one who had been an in-patient for the entire 8 months follow-up period (in addition to 3 months before baseline). 38% of the sample had made out-patient visits ranging from 2 follow-up appointments to a girl who was having thrice weekly psychoanalytic psychotherapy. 53% had had either one or two family interviews, ie a meeting at which a close relative was present. 71% had visited their GP on eating disorder matters. The number of visits to GPs ranged from 1 to 18.

It is difficult to summarise the treatment data satisfactorily because such was the variety in both mode and intensity that each case seemed to have followed an independent course. At the most severe end of the spectrum, treatment mode and frequency bore only a tenuous relationship to severity of illness. 1 or 2 of the illest patients were being managed by their family doctors alone. For example, the only treatment that a girl with 56% W/H had received in the previous 3 months was 2 visits to her GP for weighing. One patient at 72% W/H was currently being treated in a private clinic for alcoholism.

How effective any of the treatment was in effecting change remains a conundrum. The difficulty of assessing effectiveness can be illustrated by describing the experience of one patient. At the initial interview she was an in-patient. She discharged herself 3 weeks later returning to the parental home. After her school refused to have her back on medical grounds, her GP referred her to a general paediatric unit where she was put in an isolation cubicle for re-feeding. She ran away 3 days later and was picked up after a few hours by the police. Apparently, the police read her the riot act and told her how selfish she was being, wasting police time and so on. They took her home. She and her mother then worked out a joint diet plan. Her weight began to rise. What was the effective ingredient here? The in-patient stay? The isolation cubicle? The police? The diet plan? None of these?

7.5.2 Cystic Fibrosis

92% of patients had had some contact with treatment agencies in the previous 3 months and a third had been hospital in-patients at some point in this period. There was a much more obvious relationship between illness severity and hospital contact than in the AN group. 84% had attended hospital out-patients ranging from 1 to 8 appointments. 65% of the sample had had some contact with their GP on CF matters over this period.

TABLE 7.2 TREATMENT IN THE PREVIOUS 3 MONTHS % RECEIVING TREATMENT AND DAYS IN TREATMENT

	AN n = 26	CF n = 25
In-patient treatment No of days (Range)	15% 3-90	36% 1-8
Day patient treatment No of days (Range)	8% 21-26	0% -
Outpatient treatment No of visits (Range)	38% 2-30	84% 1-8
GP treatment No of visits (Range)	71% 1-18	65% 1-4

7.6 Health Status at Follow-up

7.6.1 Anorexia Nervosa

At 9 months follow-up health status data was available on 26 of the original sample of 27. There was some statistically significant improvement in health status at follow-up in the AN patients. Weight rose from a mean of 79% of that expected for height to 82%. Morgan and Russell scores rose from a mean score of 6 to a mean of 7.2 (higher scores are healthier) and the GHQ measuring general psychological distress fell from a median score of 8 to one of 3.5. These results and the appropriate statistics are summarised in Table 7.3. However, the change is clinically unimpressive.

Only 8 of the patients could be described as fully recovered in that they no longer met any of the 4 DSM-III-R criteria for anorexia nervosa. 1 of these 8 now met criteria for major depressive illness. Morgan and Russell (1975) suggest 3 categories of outcome (General Outcome Score): a "good outcome" in which body weight is within 85% of W/H, bulimia is absent and menstruation is normal; "intermediate outcome" in which weight has risen to within 85% of W/H but amenorrhoea persists and "poor outcome" where the patient is below 85% of W/H and amenorrhoea persists or where bulimic episodes are occurring on a weekly basis. Using these categories, 20% of the group had a good outcome, 31% an intermediate outcome and 50% a poor outcome.

TABLE 7.3 ANOREXIA NERVOSA PATIENTS CHANGES IN HEALTH STATUS AT FOLLOW-UP n = 26

Means	Baseline	Followup	SD_{diff}	t^1	p
% Average Weight	78.5%	82.4%	7.9	-2.50	**.02**
Morgan & Russell AOS	5.9	7.2	1.7	-3.87	**.0007**
Median				Z^2	p
GHQ	8.0	3.5		2.06	**.04**

[1] paired t test
[2] Wilcoxson's matched pairs test

As a check against rater bias (the main rater at follow-up was not blind to the objective of the study, see Section 7.1 for explanation), a sample of 15 audio tapes of the Morgan and Russell interview were also rated by an independent assessor[2]. Agreement was excellent between raters. The limits of agreement are shown in Table 7.4. Morgan and Russell scores can range from 0 to 12. The biggest mean difference between raters was 0.36 (Mental State), ie less than one point of difference and the 95% confidence intervals were all less than one point of difference.

[2] Mrs Wilma Warwick, Research Associate, University of Edinburgh

TABLE 7.4 MORGAN AND RUSSELL SCALES INTER-RATER RELIABILITY AT FOLLOW-UP MEAN DIFFERENCES BETWEEN RATERS n = 15

Scales	Mean Difference	95% limits of agreement
Food Intake	0.35	0.80 to -.1
Mental State	0.36	0.89 to -0.19
Menstrual Status	0.0	Perfect agreement
Socioeconomic	0.23	0.86 to -0.40
Psychosexual	0.04	0.41 to -0.32
Average Outcome	0.21	0.49 to -0.06

7.6.2 Cystic Fibrosis

Health status data was available for 25 of the original sample of 29 patients and is summarised in Table 7.5. In the 8 months period there was on average little change in health status. Lung function was unchanged and there was a small statistically significant but clinically negligible improvement in weight. Excluded from this analysis were the 2 patients who died. Both had been very ill at the baseline assessment. The CF group were reporting less emotional distress between baseline and follow-up (GHQ scores).

TABLE 7.5 CYSTIC FIBROSIS PATIENTS HEALTH STATUS AT FOLLOW-UP n = 25

Mean Scores	Baseline	Follow-up	SD_{Diff}	t^1	p
% Average Weight	86.0%	87.5%	3.9	-2.0	**.05**
FVC	75.9%	77.5%	14.1	0.55	.58
FEV_1	62.5%	62.6%	11.9	0.03	.97
Median				Z^2	p
GHQ	2.0	0.5		2.06	**.04**

[1] paired t test
[2] Wilcoxson's matched pairs test

7.6.3 Well sample

In the well group weight data was available for 28 out of the original 31 sample. There were no changes between baseline and follow-up, mean weights being 95% and 94% of expected respectively.

7.7 Self Report Questionnaires of Family Functioning

7.7.1 Household

The means and standard deviation of the household climate scores on the FAM and the 3 subscales of EFS: Conflict, Enmeshment and Rigidity, are presented in Table 7.6. Because of some attrition between baseline and follow-up, the means at baseline are not identical to those presented in the initial analysis (Sections 6.4.1 and 6.4.2). A 2-way MANOVA design with subject groups as the independent variable and 4 dependent variables (the 4 test scores) with repeated measures (baseline and follow-up scores) was selected for the first analysis. There was a significant effect for time (Wilks' lambda = 0.86, df 4,76, p = .02) but not for groups. Univariate analyses of the 4 tests showed that this effect was confined to the EFS Conflict Subscale (F = 5.04, df 1,79, p = .03) where families reported slightly

less conflict at follow-up. This difference between means is so small as to be clinically meaningless (Table 7.7).

These results suggest that there was little change in self report of household family functioning between baseline and follow-up for any of the 3 groups.

TABLE 7.6 SELF REPORT MEASURES: HOUSEHOLD SCORES AT BASELINE AND FOLLOW-UP

	AN n = 27		CF n = 26		Well n = 29	
	T_1	T_2	T_1	T_2	T_1	T_2
FAM						
Mean	51.6	51.0	52.5	52.2	50.8	50.8
(SD)	(5.1)	(4.7)	(4.1)	(4.2)	(5.3)	(5.7)
EFS Conflict						
Mean	52.0	49.6	52.3	51.6	51.1	49.7
(SD)	(7.8)	(6.9)	(6.8)	(7.2)	(8.3)	(8.8)
EFS Enmeshment						
Mean	51.8	49.7	50.5	51.0	52.5	53.3
(SD)	(7.4)	(7.0)	(6.9)	(7.9)	(7.0)	(9.8)
EFS Rigidity						
Mean	46.0	50.6	50.6	50.7	48.9	50.7
(SD)	(6.1)	(4.8)	(4.8)	(4.9)	(6.2)	(6.1)

T_1 = baseline T_2 = follow-up

2 way MANOVA Groups x Time (Baseline to Follow-up)

Groups	Wilks' lambda	.86; d.f. 8,152; p .16
Time	" "	.86; d.f. 4,76; **p .02**
Groups x Time	" "	.84; d.f. 8,152; p .12

TABLE 7.7 SELF REPORT MEASURES CHANGE IN HOUSEHOLD SCORES BETWEEN BASELINE AND FOLLOW-UP

Variable	d.f.	F[1]	p
FAM	1,79	0.65	.42
EFS Conflict	1,79	5.04	**.03**
EFS Rigidity	1,79	0.28	.60
EFS Enmeshment	1,79	3.62	.06

[1] ANOVA Univariate Analysis of Variance

7.7.2 Index Child

In the baseline analysis there were some differences between groups in the self report scales completed by the index child. On the EFS the AN patients were found to report slightly more enmeshment than the CF patients and CF patients reported more rigidity than the well and AN groups. In the circumstances it seemed justified to look separately at the index child scores at follow-up although these scores contribute to the household index described above. The means and standard deviations are presented in Table 7.8. The group effect was significant (Wilks' lambda = 0.74, df 8,146, p = .003) with no effect for time. Further analysis (Table 7.9) showed that this effect was present in the EFS rigidity scale only (F = 3.9, df 2,76, p = .02) and was accounted for by the CF children reporting more rigidity than the AN group (Duncan's post hoc comparison of means test AN v CF p = .04). An examination of the means shows that the reason AN patients at follow-up no longer showed more enmeshment than the CF patients was that their scores had remained stable while the CF scores shifted closer to the AN means.

TABLE 7.8 SELF REPORT MEASURES: INDEX CHILD SCORES AT BASELINE AND FOLLOW-UP

	AN n = 26		CF n = 24		Well n = 29	
	T^1	T^2	T_1	T_2	T_1	T_2
FAM						
Mean	51.6	50.0	50.7	50.7	50.6	49.3
(SD)	(9.3)	(6.7)	(5.1)	(5.2)	(8.1)	(6.9)
EFS Conflict						
Mean	52.3	47.2	47.6	48.3	50.1	48.7
(SD)	(13.2)	(10.1)	(7.2)	(7.3)	(11.4)	(11.8)
EFS Enmeshment						
Mean	54.8	54.7	48.5	51.0	52.1	51.7
(SD)	(10.2)	(11.8)	(8.7)	(8.8)	(9.7)	(11.6)
EFS Rigidity						
Mean	48.2	48.0	54.6	52.6	49.4	51.8
(SD)	(9.8)	(7.7)	(6.3)	(6.9)	(8.6)	(7.5)

T_1 = baseline T_2 = follow-up

2 way MANOVA Groups x time (Baseline to follow-up)

Groups	Wilks' lambda	.74; d.f. 8,146; p = **.003**
Time	" "	.92; d.f. 4,73; p = .22
Groups x time	" "	.84; d.f. 8,146; p = .14

TABLE 7.9 SELF REPORT MEASURES CHANGES IN THE INDEX CHILD BETWEEN BASELINE AND FOLLOW-UP

Variables	d.f.	F[1]	p
FAM	2,76	0.94	.87
EFS Conflict	2,76	0.56	57
EFS Rigidity[2]	2,76	3.88	**.02**
EFS Enmeshment	2,76	1.76	.18

[1] ANOVA Univariate Analyses of Variance

[2] Duncan's Post hoc comparison of means CF v AN p = **.04**

7.7.3 Parents and Siblings

No further analyses were carried out at follow-up on parent and sibling self report scores firstly because in the initial analyses no difference between groups had emerged and secondly at follow-up no differences were found in the household scores (to which their test scores had contributed).

7.8 Expressed Emotion

Tables 7.10 and 7.11 show the median scores for critical comments and EOI at baseline and follow-up for the AN and CF families respectively. No hostile remarks were made at follow-up. The only individual EE scores to show significant change were some reduction in the number of critical comments made by AN mothers and a lowering of EOI scores for both sets of fathers from an already low baseline. The proportion of AN households defined as high EE dropped from 52% to 43% and in CF households from 32% to 24% neither drop being statistically significant.

In the AN group there was no relationship between whether a household made a high number of critical comments at baseline and the AN health outcome on the General Outcome Score of the Morgan and Russell Scale ($r = .06$, $p = .79$). There was however a correlation of $-.37$ between high EOI at baseline and poor outcome at follow up although it just failed to reach significance ($p = .06$).

As the number of positive remarks and warmth do not form part of the EE index, they are not reported at follow-up, particularly as the inter-rater reliability score for warmth at baseline had been so weak.

TABLE 7.10 EXPRESSED EMOTION ANOREXIA NERVOSA PARENTS AT BASELINE AND FOLLOW-UP

	Baseline Med (Range)	Follow-up Med (Range)	Z^1	p
Mothers n = 21				
Critical Comments	2 (0-5)	1 (0-3)	2.18	**.03**
EOI	2 (0-4)	2 (0-4)	0.85	.39
Fathers n = 18				
Critical Comments	1 (0-4)	1 (0-3)	1.65	.10
EOI	0.5 (0-4)	0 (0-3)	2.10	**.04**
			χ^2	p
High EE household[2]	52%	43%	0.13	.72

[1] Wilcoxson's Matched Pairs Test
[2] McNemar's χ^2 for matched proportions

TABLE 7.11 EXPRESSED EMOTION BASELINE TO FOLLOW-UP CYSTIC FIBROSIS

	Baseline Med (Range)	Follow-up Med (Range)	Z^1	p
Mothers n = 25				
Critical Comments	1 (0-10)	1 (0-5)	0.33	.74
EOI	2 (0-5)	2 (0-4)	0.82	.41
Fathers n = 16				
Critical Comments	1 (0-8)	1 (0-7)	0.04	.96
EOI	1 (0-4	0 (0-2)	2.52	**.01**
			χ^2	p
High EE household[2]	32%	24%	0.25	.61

[1] Wilcoxson's Matched Pairs Test
[2] McNemar's χ^2 for matched proportions

7.9 Problem Solving Task

The results for 3 measures derived from the problem solving task at baseline and follow-up are summarised in Tables 7.12 and 7.13. There were no significant shifts in the amount of overprotective behaviour or signs of weak generation boundaries observed at baseline and follow-up for either

the AN or the CF families. In the CF group there were no examples at all of weak generation boundaries in the CF group at follow-up.

Problem solving ability was dichotomised to facilitate statistical analysis. Good and intermediate ability categories were merged to form one category while poor problem solving skills formed the other category. At follow-up 26% of the AN group showed poor skills compared to 58% at baseline, a difference which did not reach significance. In the CF group 43% had shown poor skills at baseline compared to 14% at follow-up which was a significant drop (McNemar's $\chi^2 = 4.9$, $p < .03$).

These findings could indicate a genuine improvement in skills. An alternative explanation is that the groups were more practised at the task, having already rehearsed one of the problems at baseline. Another factor is that problem 2 at follow-up was to "plan a menu" rather than discuss a family problem. This may have been easier for the CF families than the AN families and explain why the CF families appear to have improved more.

TABLE 7.12 PROBLEM SOLVING TASK CHANGE AT FOLLOW-UP ANOREXIA NERVOSA n = 19

	Baseline Med (Range)	Follow-up Med (Range)	Z^1	p
Overprotection	5 (0-17)	4 (0-16)	0.98	.33
Weak Boundaries	3 (1-2)	2 (1-3)	0.81	.42
% Poor Problem Solving Skills	58%	26%	χ^{22} 0.44	p .50

[1] Wilcoxson's matched pairs test
[2] McNemar's χ^2 for matched proportions

TABLE 7.13 PROBLEM SOLVING TASK CHANGE AT FOLLOW-UP CYSTIC FIBROSIS n = 21

	Baseline Med (Range)	Follow-up Med (Range)	Z^1	p
Overprotection	1 (0-16)	3 (0-9)	0.36	.72
Weak Boundaries	0 (0-2)	0 (0)	-	-
% Poor Problem Solving Skills	43%	14%	χ^{22} 4.9	p **<.03**

[1] Wilcoxson's matched pairs test
[2] McNemar's χ^2 for matched proportions

7.10 Change in Health Status and Change in Household Climate

To examine the relationship between change in severity of illness between baseline and follow-up and changes in household climate over the same period, correlations between illness change scores and household climate change scores were calculated. The illness variables for CF patients were percentage of weight expected for height (%W/H) and the composite score derived from weight and lung function (described in Section 6.9). The illness variables for AN patients were % W/H and the Morgan and Russell Average Outcome Score. The 3 household climate variables were derived from the factor loadings of the principal component analysis described in Section 6.8. These were negative family self evaluation (Factor I), observed enmeshment and overprotection (Factor II) and observed criticism and poor problem solving skills (Factor III). The correlations are summarised in Tables 7.14 and 7.15. The only significant correlation in the CF group was between illness severity and factor III ($r = .47$, $p < .03$). What that suggests is that as health deteriorates, families become more critical and worse at solving problems. In the AN group a correlation between Factor II, observed enmeshment and overinvolvement and weight, just failed to reach significance ($r = .40$, $p < .09$). Here a rise in weight suggested a lessening of enmeshment and overprotection. Calculating confidence intervals for these correlations dampens much enthusiasm for these findings. The 95% confidence intervals for CF severity and Factor II yields a correlation range from .75 to .05. For AN Factor II and weight the confidence intervals range

from .72 to –.07. Removing 1 outlier case from the CF group reduces the correlation between illness severity and Factor III to .33 (p = .15).

There were no significant differences between correlations for CF families and correlations for AN families.

TABLE 7.14 CORRELATIONS BETWEEN CHANGES IN HEALTH STATUS AND CHANGE IN FAMILY FUNCTIONING CF FAMILIES n = 21

	% W/H	CF severity
Factor I	.30	.21
Factor II	.18	.20
Factor III	.06	.47[1]

[1] p = **<.03**

TABLE 7.15 CORRELATIONS BETWEEN CHANGES IN HEALTH STATUS AND CHANGE IN FAMILY FUNCTIONING AN FAMILIES n = 19

	%W/H	Morgan & Russell
Factor I	-.02	-.19
Factor II	.40[1]	.01
Factor III	.06	.04

[1] p < .09

CHAPTER 8: A DESCRIPTIVE ACCOUNT

For the last 2 chapters the reader has had to digest a somewhat dry diet of facts and figures which afford little flavour of the experience of families in the study. To redress the balance, this chapter contains a descriptive account of some of the families and some of their experiences. In the first half of the chapter, case vignettes will be given of some of the families in the study. In the second half of some of the experiences of families will be described, looking in particular at the differences between the AN and CF families.

A statistical focus on the mean has obscured the wide variety of households and household styles that were encountered during the study. Happy families were no more alike than unhappy ones and were to be found in all 3 groups.

The shortcomings in one area of family functioning could often be mitigated by strengths in other areas so that quite different styles of family could function well. For example, there was a particularly boisterous and chaotic family (Case 304), much given to shouting, arguments and verbal abuse. Hence they had high scores on the Conflict scale and critical comments. But they were also tremendous fun, frank, warm, forgiving and tolerant of individual peccadillos. Their cheery household seemed to be a mecca for various lame ducks and oddball teenagers, 2 of whom had to be cleared from the room before the interview could begin. By contrast, another family (Case 221) had a very quiet orderly lifestyle punctuated by regular meals and favourite TV programmes. Father and son were both computer enthusiasts and spent most of their free time communicating with the flickering screen. Yet this household which at first sight appeared so emotionally repressed and rigid seemed to be functioning well. Their strengths seemed to be a certain dogged persistence and unflappability in times of crisis. The low level of "white noise" in the home allowed them to tune in to quite faint frequencies. There had been family crises in the past

and the family had dealt with them with great sensitivity and flexibility. The son was a CF patient and the parents had made an excellent job of normalising his childhood without compromising his health care.

Before undertaking this study I had no experience of interviewing well families and was immediately struck by observing many behaviours that would be deemed pathological if witnessed in the psychiatric clinic, yet seemed to have no effect on the usual individual parameters of psychological well-being or social functioning. The 2 families who most closely matched Minuchin's "psychosomatic family" were in the well group.

8.1 Case Vignettes

Cases were chosen not because they were representative but because they illustrated some of the family characteristics of interest to the study. All names are false.

8.1.1 A Family with Negative Affective Style (Case 203)

At the baseline interview Anne was 21, had just finished higher education and was living at home with her parents. Her closest friend was her brother who had now left home to live with a girlfriend.

CF had been diagnosed when she was 3 months old. Until Anne was 11 her health was good, maintained by regular physiotherapy and pancreatic enzymes. She was keen on sports, winning school cups for games and was known as an extrovert. As she got older and lung function began to deteriorate, according to her mother she changed from being outgoing to being extremely private and resentful of the limitations of her illness.

At interview she had no psychiatric illness but admitted to having short spells of black anger about her condition. Had the dice been cast differently, she would have liked to be tramping through the rain forests of

the Amazon. As it was she was confined by her illness to nothing more strenuous than short walks with her dog and rides in her car and was trapped with parents with whom she felt she had little in common.

Anne was meticulous over the management of CF. She resented any interference from her mother who persisted in monitoring her daughter's health and attempting to control it. For example, the mother would telephone the hospital to make appointments for her daughter who promptly cancelled them to remake her own appointments.

Mother was low in warmth and high in criticism on the Expressed Emotion index, critical not only of her daughter but of most topics we discussed. The affective style of both mother and daughter was negative with much criticism, justification, sarcasm and shouting on both sides and no positive solutions or acceptance. Discussion of 2 family problems during the 10 minute task led to an escalation of the problems rather than a resolution. Father was silent and uncommunicative. At follow-up, he refused to be re-interviewed but gave no reason for this.

8.1.2 An Overinvolved Family (Case 222)

Brian was 22 years old but still attending a paediatric clinic for CF patients. When I queried this, his mother told me that the clinic loved him so much they couldn't bear to let him go. However, he had made the move by the time of the follow up interview. At interview he looked and seemed much younger than his years, an uncomplicated person with a placid temperament and very low GHQ score. His lung function was about average for the CF group although his weight was a little below average. He had had IV antibiotics in the previous 3 months and a total of 5 admissions to hospital to bring infection under control. He had recently been chosen to go on an expenses paid holiday to Canada to a special summer camp for CF patients.

Brian's mother was one of the handful of mothers with very high overinvolvement scores on the Expressed Emotion index. She also had a high GHQ score (20) although no mental health history. There were many examples of self sacrificing and overprotective behaviour. She worked outside the home, not in order to give herself a break but in order to buy Brian the little extras she felt he needed. Brian had qualified in a trade that had been selected for him by his mother but there was no demand for that trade in the area of high unemployment in which the family lived. Mother was unwilling for Brian to move away to a town where he might be able to find work. The emotional tone of the CFI was one of exaggerated emotional response and dramatisation. Brian's father spoke for no more than 5% of the interview.

However, the family were not enmeshed. Brian showed none of the overprotective behaviour towards his mother that she showed to him. The boundaries between generations were strong with Brian clearly in the child role, albeit a role more appropriate for a 12 year old boy than a 22 year old man. Brian seemed to recognise the inappropriateness of his mother's behaviour. One of the problems discussed in the problem solving task was his feeling that his mother pampered him too much. However, little progress was made during the discussion and Brian showed no real inclination for independence.

8.1.3 A Family with Physical Burden (Case 218)

Clare was one of the illest CF children in the study. She was 20 years old. She was diagnosed as having CF at the age of 4 but it was not until she was 15 that she had her first major infection and admission to hospital. Her weight was 71% of expected, her lung function was too low to be measured but was estimated to be FVC 23% and FEV_1 15%. She was too ill to attend out-patients and was being cared for at home by her mother who had given up work 2 years earlier to take on this task. At the time of interview she had been on the heart-lung transplant list for 2 years and had

just been moved to the priority list. Getting on to the list had involved a 2 week assessment in London. Father describes the strain.

> "It was pretty harrowing for Clare, you know. It was all the tests she had to go through ... obviously they don't want to carry out such an operation on somebody if they feel the family support is not there, you know. So I felt we were under a microscope quite a bit as well."

At the time of interview, the family were in the process of having a bedroom and bathroom built on to the house downstairs but in the meantime Clare spent most of her time in her bedroom. To improve her blood gases, she had been advised to use her oxygen cylinder most of the time even if she felt she did not need it. Her mother described her average day.

> "She gets up at about 9 and then she uses her nebuliser and then she gets her physio [from mother]. And then its breakfast about half past ten and then she uses her nebuliser about half twelve and then she has lunch just after one. It's a shorter session at lunchtime. And she's doing a Higher course in Economics just now, so she does some studying in the afternoons ... then it's physio again after three and it's tea-time."

The burden on the parents was reflected in their Impact on Family Scale scores, 66 for mother (CF group: Mean (SD) 54.0 (7.0)) and 58 for father (CF group: Mean (SD) 49.5 (6.6)).

However, in spite of the burden, the family and its individual members had good psychological functioning. All the household had GHQ scores below 5 and no mental health history. Their FAM scores and EFS scores were at the normal sample mean. Neither parent showed emotional overinvolvement. The family had adapted to the illness acknowledging the difficulties but not being consumed by them. For example, Clare's mother was sorry that she had had to leave her job but conceptualised her present responsibilities of looking after Clare as her new job which she found rewarding. She still went out to a Saturday morning job. In spite of Clare's

physical dependence on her mother, the mother had not infantalised her and recognised her adult needs. For example, at weekends several of Clare's friends would come round in the evening. The parents made a point of going out themselves partly, father explained, because no young person wants their parents breathing down their neck and partly because they couldn't stand the heavy metal music. On the problem solving task the family discussed one of the most common family problems: the tidiness of Clare's bedroom. Clare's immediate reaction to the problem was one of self justification but after her mother persisted with uncritical self disclosure, a frank discussion followed with evidence of seeking clarification, positive solutions and acceptance. The discussion ended with Clare saying that she was going to do nothing to change the arrangement of her CF paraphernalia but that she would try and tidy up some of her books and records. Father and Clare agreed to explore whether an extra cupboard could be built into the room. This movement towards resolution was scored as good problem solving skills. There was no evidence of enmeshment and generation boundaries were clear.

On interview Clare's psychological well-being was remarkable. Her disposition was relaxed and cheerful. She expressed interest in the research project and wanted to know all about the AN sufferers and the Glasgow CF patients. There was no evidence of denial of illness. She understood her condition well and had no illusion about her disability. Clare had died before the follow-up interview could take place. Her mother commented "she was everything any parent would want in a daughter, she was marvellous".

8.1.4 A "Psychosomatic Family" (Case 103)

Diana was 16 years old. She had had AN for 4 years and at the time of baseline interview had been an in-patient for several weeks. Her weight was 56% W/H. The household consisted of mother, father, Diana and her

sister who was 18 and, although very jealous of the attention that Diana had obtained, seemed problem free herself.

When interviewing the parents alone, they told the history in the rather detached resigned way of people who had done this many times before - showing little distress or emotion. The family had a catalogue of what they felt to be disastrous experiences of seeking treatment and getting help. Father at the time of baseline interview had a depressive illness which may explain some of his flattened affect. The family described themselves as normal on the FAM scales and EFS scales. They made few criticisms of their daughter and would have obtained a low over-involvement score were it not for the fact that the history told of much self sacrificing behaviour in pursuit of their daughter's health. The family had been living abroad when the illness began and at one point had lived apart for a year with father and well daughter remaining abroad while mother and AN child returned to the UK to seek treatment. The parents eventually decided to return permanently to the UK in order to support their sick child and to do this father had resigned from his job and was currently unemployed.

After the Camberwell Family Interview, the daughter entered the room for the family discussion that preceded the problem solving task. The atmosphere immediately changed from one of detached resignation to one of heightened concern. Father had earlier described the strain of having family meals round the TV with everybody on edge and feeling like jelly. This was the mood of the problem solving task. The parents adopted a soft voiced style as though any wrong footedness would spell disaster.

The first problem to be discussed had been raised by Diana in her cue elicition interview. She had expressed the view that sometimes her parents worried about her too much and that she was fed up with them asking her how things were going. On being asked to discuss this, Diana immediately tried to diffuse the problem by saying that it was only natural for them to be concerned, that all parents worry about their children. Mother

immediately agreed with her daughter but did ask her daughter for clarification. Diana then denied that there was a problem. After a long silence, the daughter said that she worried a great deal about her parents and their problems, displaying both overprotective behaviour and blurred generational boundaries. Diana then invited nurturing behaviour by saying how guilty she felt because she was being so well looked after and they were not. The discussion ended with father saying "we'll be together sometime, in the end". By the end of the discussion, nothing of substance had been discussed and nothing resolved. The family scored 10 on observed enmeshment/overprotection (AN group: Median (Range) 2.5 (0-17)).

8.1.5 A "Hilde Bruch" Family (Case 314)

Edward's family lived in a beautiful home in a highly desirable area of the city. The house had an atmosphere of civilised calm: flowers in the hall, chamber music in the background.

Edward was the youngest of 3 children, the others having left to follow professional careers. Father was a successful business man and mother was deeply involved in charity work. It took some effort to find an evening when all 3 members would be free for interview, such was the busyness of their lives.

Edward was 18 at the time of interview. He had obtained high grades at GCSE and had a high profile in his private school, apparently successful at whatever he turned his hand to. He had a high GHQ score (26) but on interview was not depressed and did not have an anxiety disorder. He was however a troubled young man, pre-occupied with the state of the world, the meaning of life and the existence of God. He also was troubled by what he saw as the tremendous academic pressure he was under from his parents. He complained that if he stopped working for 10 minutes they would "go on at him".

The parents seemed to be quite unaware of Edward's inner turmoil. Their main complaint about him was that he had stopped working and was unlikely to obtain the Cambridge place that he was destined for. It was clear that the parents had high expectations for Edward and a wish to control his future although they presented a facade of insouciance. For example, mother said "we don't mind whether Edward goes to university or not, it's up to him" but later father said "all my family have been to Cambridge and Edward could get there too if only he tried". Mother later said "we're quite open about Edward's career plans but we think he would make an excellent lawyer".

On the Expressed Emotion index both parents made critical comments and were low in warmth. They were low on emotional overinvolvement and the family showed no enmeshment during the problem solving task. They were able to reach resolution in discussing one problem but remained in conflict on the other.

At the follow-up interview Edward had failed to get sufficient grades to secure his Cambridge place. A pall of disappointment hung over the household and a conflict had arisen as to whether Edward should go to a lesser institution: "not a real university in my opinion [father]" or retake his exams. The FAM scores had risen to the dysfunctional range.

This family was in the well group.

8.2 Parental Experience of Treatment

8.2.1 Seeking Treatment

One of the main distinctions between the CF and AN families was the gulf between the groups in their experience of getting treatment for their sick child.

Many CF parents had had difficulty in establishing the diagnosis. They were of course describing events that had taken place many years ago (Median 17 years) at a time when there was less awareness of CF among both general practitioners and the general public and before the genetic advances that made diagnosis unequivocal. The main complaint was that they had been looking after a child who was failing to thrive but that the family doctor dismissed their concerns with the explanation that they were over-anxious or inexperienced.

By contrast, AN parents had no difficulty in getting a diagnosis established. In most cases parents had correctly identified their child's problem before this was independently confirmed. The exception to this were the families of the 2 AN boys in the study, both of whom underwent extensive physical investigation before a psychiatric explanation was entertained.

A common complaint from AN families was that there were plenty of experts willing to make a diagnosis and an assessment but far fewer who were able to offer any treatment and fewer still in which treatment appeared to be effective. Most AN mothers felt that the onus of seeking help fell squarely on their shoulders. This was partly due to denial of problem by their child, partly to uninformed general practitioners and partly to a lack of appropriate resources in their areas.

Case 111 serves as a rather extreme example of this problem. At the time of baseline interview, a total of 11 different referrals had been made (all initiated by mother rather than patient). Initial referral to the family doctor (who confessed to total ignorance of AN) was followed by a 3 week admission to a general hospital for refeeding. Then followed 3 years at home without treatment but with weight at 75% ABW. When weight began to drop further, the patient's mother sought a private psychiatric consultation at which the diagnosis was reconfirmed. The patient then saw a hypnotherapist for 9 months with no change. He then referred her to someone in his "magic circle", a dental hypnotist who saw her 4 times "to

build up a picture" but offered no treatment. Then followed 5 weeks in an NHS acute psychiatric ward (this admission was organised by the original private consultant) during which the patient lost further weight.

She then saw a private psychologist who saw her 4 times to "build up a case". Her GP then referred her to another psychiatric hospital where a registrar decided to instigate a strict behaviour modification regime (isolation room with mattress on floor and disconnected washbasin), work which neither he nor the nursing staff had ever carried out before. The patient discharged herself after an incident in which it was alleged that 2 nurses mocked and jeered at her and "forgot" to bring her her meals. After reading a newspaper article, the mother wrote to a well known AN expert who finally directed her to a specialist centre where she was offered adequate treatment.

The drastic reduction in in-patient beds in recent years had led to NHS hospital admissions being restricted by and large to psychotic patients, alcoholic and drug dependent patients and patients with severe personality disorder (Wing, 1989). Furthermore, admission rates are strongly related to social disadvantage (Jarman, 1983, 1984). An AN mother bemoans the proletarian atmosphere of her local hospital.

> "It was the best they could do, it was all about funding and everything. I quite appreciate that but it was totally inadequate and totally inappropriate. She was in a ward with other people who had all sorts of problems, one of whom spat all the time on the floors, on the table, everywhere, and for somebody with an eating disorder who finds it difficult to take food, it was just dreadful ... The staff did their best but again a lot of them had no knowledge of the illness at all and told that they didn't know anything about it which really didn't do a lot for her confidence."
>
> (Case 127)

By contrast, a mother who had a good experience of seeking treatment had taken the problem to her family doctor who had skilfully engaged her daughter and seen her for a 4 week trial period. When that failed, he had

referred her to a specialist centre and continued to support both mother and patient during the wait for a first appointment (Case 109).

8.2.2 Parental Blame

Intertwined with the difficulties that many AN parents had in getting what they considered to be adequate treatment for their child was the finding that attitudes towards parents were often perceived as negative. Parents were asked whether a hospital or clinic had ever made them feel that problems arising from their child's disorder was their fault. 95% of CF fathers and 57% of AN fathers answered "no", the difference between proportions being statistically significant ($p = .009$). 88% of CF mothers compared to 60% of AN mothers answered "no", also statistically significant ($p = .03$). AN parents were more likely to answer "yes" if they had been excluded from care or if they had experienced family therapy.

8.2.3 Parental Involvement in Treatment

The most negative responses came from AN parents who felt that they had been excluded from care.

> " ... the consultant flatly refused to speak to us. Nobody would speak to us because she was his patient and we were kept very much on the outside which was really terrifying ... you saw you daughter wilting away in front of your very eyes and you had nobody to turn to ... you read about these things but you don't get the full impact for something until you are actually involved."
>
> (Case 104)

"We went to our own GP who had never ever treated anyone with anorexia before but he put us on to the local psychiatrist ... Well it was a complete disaster ... First of all the psychiatrist wouldn't talk to me at all and did talk to S [the daughter] but we found that they weren't offering her anything at all and all I could see was the weight coming off ... They transferred us to another psychiatrist and this one was worse. Again he wouldn't speak to me at all. I just couldn't get in contact with anybody."

(Case 106)

Both these mothers were later referred to another treatment agency and were able to be much more positive.

" ...One of the therapists was very good for P [the daughter] and for us and gave us the support we needed. And I really cannot speak more highly of the place."

(Case 104)

"It's not only the patient who needs help, it's the family and I honestly am totally amazed that we are a year and a half into this and it has taken a year and a half for me to feel that I have somebody if a crisis happened ... that I could get hold of or have five minutes to talk to on my own."

(Case 106)

The one group of parents who did not feel excluded were the 7 families whose child was a private patient, some of whom had had shifted to the private sector because of earlier negative NHS experience. They had all been given an opportunity to "tell their story" and had had their questions answered. In all but one case fees were being paid by either the parents or their health insurance rather than by the patient herself, an arrangement which may serve, probably unwittingly, to alter the dynamics of attitudes to the family.

AN parents who felt excluded wished for 2 kinds of help: practical advice and emotional support (see below).

It has been acknowledged that CF parents often know more about the illness than some of their medical advisors (Report of the Royal College of Physicians, 1990). A CF mother from a working class background (Case 213) told of a recent experience. Her daughter (aged 18) was not responding to oral antibiotics and mother organised admittance to hospital for IV antibiotics. However, once in the hospital, the junior doctor was not prepared to set up the drip without clearance from the consultant in charge who was out of the country. Then the pharmacy announced that they were out of the child's oral antibiotics and asked mother to bring in supplies from home. Mother and daughter decided that it was better to be at home than in hospital in these circumstances and against advice daughter discharged herself. The result of this episode was an apology from the consultant. Some of the AN parents were also well informed about the illness but they felt that their knowledge was seen as threatening to the treatment agencies. If unilateral action was taken by AN families, their behaviour tended to be construed as "acting out", "refusal to engage", "toxic parenting" rather than as an occasion for apology.

As CF patients reached adolescence, the expectation was that they would take over responsibility for their care from their parents. Adolescence also heralded the move from paediatric units where it was customary for the child to be examined in the presence of parents to adult units where patients were seen alone. There was recognition in both Glasgow and Edinburgh that there was a risk that parents might feel rejected or excluded from care and the transfer was planned with this in mind. For example, the physician for adults might first see the child and parents in the paediatric clinic or a liaison doctor well known to families from the paediatric clinic saw the parents at the adult clinic while their child was seeing the adult physician. Only 2 CF families had felt excluded from care. In one case, the mother was high on emotional overinvolvement and found it difficult to accept the growing independence of her child. In the second case, the mother mourned the loss of a favourite paediatrician and could not adjust to the different personality of the adult chest physician.

The collaboration of CF parents with the medical teams in joint care of the sick child had served to empower the CF families in a way that was quite absent from the AN families.

There was, however, one area in which the CF families felt ineffective: those involved in waiting for a heart-lung transplant. While the families had nothing but praise for the heart-lung assessment teams, the wait for suitable organs was a lottery out of their control and a source of considerable anxiety. There was a requirement that they should not go anywhere without carrying a bleep alarm that could summon them unexpectedly to the far end of the country for the operation . At that time there were no facilities for heart-lung transplants in Scotland. Whitehead et al (1991a) found high levels of psychological dysfunction in the families of young CF patients awaiting transplant.

8.2.4 Practical Advice

There was a wish for didactic information about AN and for practical advice on matters such as what was the appropriate role for the mother over weight restoration. Should she be involved in the preparation and consumption of food or should she opt out? Where advice had been given, it was often conflicting. One agency has strongly advised a family (Case 103) to force their child to eat using physical restraint if necessary. Two years later, a different agency advised them strongly never to interfere in eating at all. Neither approach appears to influence the health status of their very ill child.

By contrast the CF families, particularly in the early years, had received much didactic advice from physiotherapists, dieticians, health visitors, the implementation of which resulted in real improvement in the health status of their child. Over the years the advice had sometimes changed. For example, 20 years ago patients were advised to sleep in mist tents. The ineffectiveness of this regime led to its discontinuity but blame for this fell

on the tents rather than parental supervision of the tents. For the AN families failure of a treatment and change of advice often seemed connected with apportioning blame.

8.2.5 Support Groups

35% of AN parents and 65% of CF parents had attended some sort of parental group. AN families had attended nurse led non-directive groups designed to give them emotional support. Half were for mixed psychiatric populations and half for AN families only. No AN family had found the groups helpful although 2 families derived some comfort from finding that other families had problems much worse than their own. No parents had attended the kind of structured goal directed groups run by the Toronto Eating Disorders Clinic. Lack of structure and apparent lack of objectives in groups were found particularly stressful.

> "We were expected to raise topics for discussion. Nobody liked to do this because the nurse would then lay into you. So sometimes we all just sat there for 10 minutes, 20 minutes, in embarrassed silence ... I asked if I could leave early to catch the last ferry home. The nurse interpreted this to the group as me never putting my daughter first. If we had stayed, we would have had to spend the night in a hotel and get the ferry next morning. We couldn't afford it."
>
> (Case 103)

The CF families had attended group meetings run by the Cystic Fibrosis Trust which tended to be topic focused didactic evenings, eg "heart-lung transplants", or "advances in genetics". These evenings had a mixed reception. Some found them informative and helpful, others preferred to receive information directly from their physician. Some families saw the CF Trust as a middle class organisation and felt excluded from the meetings. A working class father explains:

> "It was more like a sort of snobbish thing as opposed to being more informal ... you were sort of embarrassed when you went to these places because it was the doctors and professionals and different people and you didn't feel like asking questions."
>
> (Case 206)

Twenty years ago Cull (1974) had found a similar reaction from working class households to Trust meetings in Scotland.

No CF parent had attended any groups offering emotional support and none expressed any wish for help of that kind.

8.2.6 Family Meetings

85% of AN families had at some time had a family meeting, ie a meeting with patient, parent(s) and therapist present. This was usually an assessment interview and the therapist was most likely to be the family doctor. Nine families had received family therapy, loosely defined here as at least 2 family meetings with a therapeutic orientation. Three different psychiatric units were involved. Eight of the nine families had found family therapy a negative experience and that experience often led to them withdrawing from treatment.

> "R [husband] and I used to come home really drained. They [the meetings] were always bad. They had the screen ... the one way screen ... you had to speak about things you preferred not to talk about ... and somehow you came home and maybe were upset for a couple of days afterwards ... I think you feel guilty too ... I think some of the questions were meant to, maybe not, make you feel terribly guilty ... after you've answered it all and gone through it all, you think maybe it was my fault or something."
>
> (Case 112)

At 8 month follow-up, this family's AN daughter remained an in-patient with no weight change. A father in a different family recalled

> "The last time we went, it wasn't just the person who was interviewing us ... I got slightly annoyed because I said "How can you expect a teenage girl to open up when there's 4 people, strangers, sitting behind a screen ... it was going reasonably and then he got called out and they obviously told him to take a much heavier line with us and he started laying down the law with us ... I got the impression very much of a corrective institution almost ... you will do this, you will do that ... it sort of dented my confidence about fathering."
>
> (Case 114)

One family's negative reaction was qualified:

> "When you came out of the first interview, you felt really drained ... it was really quite bad ... My comment as I was leaving was 'I don't know what you're [the therapist] doing but I feel as if I've been through 15 rounds with Cassius Clay: battered'. It effected the little boy [sibling] quite badly. He refused to go back ... it was like a horrible dose of medicine that you take to do you good."
>
> (Case 121)

This family continued to work with the treatment agency and at the follow-up interview the patient had recovered.

Another mother found the family meetings helpful because they were the only forum in which her daughter spoke to her (Case 119).

These quotations are a painful reminder of Macleod's (1981) sharp criticism of certain family therapy procedures (in particular Minuchin's approach) which she found particularly brutal and distasteful.

> "As far as I can gather from the transcripts of these sessions, every member of the family, whether adult or not, is being treated as a rather unintelligent child who can never hope to understand the mysteries of the hierarchic profession whose skills are being utilised for the family's own good, and must therefore not be questioned ... I kept asking myself, how can these people stand to be treated in such a manipulative and condescending manner? The answer becomes clear soon enough: they would do anything to help their daughter/sister to save her from imminent death and being helpless to do so themselves, they had placed all their faith in what seemed to be the only hope left for them."
>
> Macleod, 1981

No CF family had experienced family therapy.

8.2.7 Individual Support

Only 3 CF mothers expressed any wish for individual support. One had a major depressive illness for which she was currently receiving psychiatric help. The second had long standing agoraphobia and the third had had 2 episodes of minor depressive disorder for which her family doctor had treated her. A more typical response was that given by the mother of Case 222. Describing a recent health crisis of her son which it was thought he would not survive, she said

> "I was anxious and upset and couldn't sleep. But I knew I wasn't ill. I knew why I was like this. I'd just rather get through it with my family and friends."
>
> (Case 222)

Six AN mothers commented that their family doctor had been a wonderful source of support but that the doctor was handicapped because he didn't know any more than they did what was actually going on in the treatment of their child.

What was wanted was some direct contact at some point with the person treating their child and occasionally, but much less rarely, some support for themselves by a separate therapist.

8.3 The Burden of Illness

A review of the literature in Chapter 1 found that psychological burden was more of a stress factor to the parents of sick children than physical burden. For many families in this study, this was true and was one of the distinguishing features between AN and CF families.

8.3.1 Individual Psychopathology

The CF families were for the most part caring for a child who was as psychologically fit as the well children in the study. Not only was there a current lack of neurosis but for most CF young people a history free of psychiatric disorder. Furthermore, CF children had good social functioning. 38% of them currently had a girlfriend/boyfriend and many more had a gang of friends whereas none of the AN group currently had a girlfriend/boyfriend.

The combination of emotional stability, physical ordeal and early mortality seems to put CF patients high on the list of the deserving sick in both the eye of the public and the medical profession. There were many illustrations of this. One CF mother (Case 206) described how her CF child was showered with cards and presents on her birthday and was keenly aware that this did not happen to her well child. Another (Case 208) told of the occasion when the family made their final visit to their London CF clinic before moving to Scotland. The child was invited to play his cornet before the entire CF team and left laden with presents. Two CF patients had been on trips to Disneyland paid for by an airline in a publicity exercise. Six had been to summer camp in Canada. None of the AN patients had received positive affirmation of this kind.

8.3.2 Household Authority

The changing role of women seems to have done little to shift their position in the home. In all 87 households visited, the mother was seen as the "chief executive" of the household. Other family members would occasionally help but their role was that of assistant rather than manager. In only one household had mother delegated responsibility for cooking and food shopping to another (her husband).

In their efforts to gain control, many AN patients had succeeded in undermining the authority of the household by enforcing exacting conditions over the management of their food. One father (Case 103) complained that he could never just go and have a snack or a piece of toast if he felt like it. The family had to have "proper meals" sitting down. Failure to do so led to his daughter rushing to her room and refusing to eat. A mother (Case 106) described leaving for work each morning having prepared 4 calorie controlled meals for her daughter, each in its own labelled foil dish and arranged in a particular configuration on the kitchen table. Another mother (Case 111) described how her daughter came home from work at midday each day to make lunch. This was to ensure that her mother ate exactly the same quantity of food as she did. Another family (Case 112) explained how they were not allowed into their kitchen between 4.30 and 6.10 each day while their daughter prepared her own food. Entry to the kitchen on whatever pretext would result in the daughter running out of the house and several hours later telephoning from a distant call box asking to be collected. Most of these families realised the inappropriateness of the position they found themselves in but felt powerless to do anything about it.

The CF families were also having to make special arrangements over feeding but in no case was food used as a psychological weapon. Many of the CF patients needed a very high calorific intake to maintain their nutritional status. Although it was a financial drain on some of the

families, most mothers, particularly in the case of sons, took some pride in the need to "feed the child up". Some households managed the need for extra calories by serving extra large portions, others by the child having an extra meal per day. In no case did the family feel a need to adapt their food pattern to accommodate the CF child's needs.

Three CF mothers of very sick children reported concern over the food intake of their children which was less than recommended levels. Acute respiratory exacerbations were common causes of restricted oral intake (see Section 3.4.3). In one household this led to unproductive arguments over how much the child should eat. In the others mothers accepted the situation with resignation.

8.3.3 Bizarre Behaviour

Behaviours that families considered bizarre were a considerable source of psychological stress to AN parents. The rigidity of food preparation and mealtimes has already been described. Obsessional time keeping was a source of distress. One patient (Case 112) insisted on going to bed at 12.17 precisely and on always being up before her mother in the morning. After her mother finally refused to postpone the start of her day to accommodate her daughter, the patient started getting up at 4 am to start her aerobic exercises.

Restless behaviour was another irritant to parents. A mother explains:

> "It's hard to explain somebody with such obsessional behaviour - having to live with somebody like that. You feel as if you want to grab hold of him and stuff food down his throat or you feel you want to kill him because he's obviously doing a good job himself and you want to put him out of his misery. There would be all night sessions in here. He'd be cooking all night in obsessional cleanliness. I mean if he touched something, he'd wash his hands, so the water would be dripping all night. The television would be on. He was just almost impossible to live with. Meantime he would still be doing his weights, goose-stepping up and down the corridors and running up and down the stairs and, when you've had a day at work, you want to come home and relax ... it just never let up and my husband went and worked away for a while ... he was very patient seeing it was my second husband and no real relative. He couldn't face coming back here to live and that's why we split up."
>
> (Case 125)

> "By this stage C. was maybe only having a lettuce leaf or a couple of dried apricots during the day and she had become very violent, aggressive and was threatening to commit suicide ... I phoned my GP once or twice again because we had these crises and she just sort of said she couldn't do anything and I was to call the police ... She did things like brandishing a knife, not at me but at herself ... she broke cups and plates, kicked at doors and it seemed that nobody was willing to do anything."
>
> (Case 124)

Although some parents felt that they had learned to understand the bizarre behaviour, few felt that they had effective repertoires to deal with it. There was a strong sense of their ineffectiveness.

8.3.4 Parental Ineffectiveness

Lawrence (1979) has described the "control paradox" of AN: the powerful control that patients exert in the area of food and weight and their own experience of themselves as utterly out of control on the other. Parents witness the paradox as a breakdown of household authority and distressed and bizarre behaviour and are often defeated by it.

There seemed to be 2 main causes of parental ineffectiveness in AN households: the neurotic difficulties presented by their child which contrasted so sharply with the average CF child and the unhelpful reception that many of them had experienced from treatment agencies unlike the collaborative relationships found by the CF parents with their health care workers.

The feeling of parental ineffectiveness was what I believe led to several parents describing that at the height of the illness they were "walking on eggshells", afraid to say the wrong thing, do the wrong thing lest there should be either an emotional outburst or a reduction in food intake. This fear of stirring things up may explain why 56% of AN families performed poorly on the problem solving task compared to only 33% of the CF group and 32% of the well group.

CHAPTER 9: THE SCHOOLS PROJECT

9.1 Introduction

In addition to the main study, a supplementary project was carried out in 2 secondary schools. This had 3 main objectives:

(1) To provide normative data for the Family Assessment Measure (Skinner, 1983), a self report measure of family functioning that has previously been used in North America only.

(2) To provide normative data for a life events schedule focusing on the stress events common to teenage and young adult households.

(3) To develop a self report scale (Edinburgh Family Scale) that would operationalise Minuchin's "psychosomatic family" dimensions: "enmeshment", "overprotection", "rigidity" and "lack of conflict resolution".

9.2 Procedure - School A

9.2.1 Subjects

School A was a private urban secondary school for girls (school role 584). Social class of the pupils was not directly measured but the majority of girls would be in classes I and II with a few in IIIa. This mirrors fairly closely the social background of the Anorexia Nervosa group in the main study. A class (mean size 20 pupils) in each of 5 school years was selected for inclusion in the study, the selection method being the pragmatic one of convenient timetabling. The classes were of mixed ability and not organised according to subject choices. Ages of the pupils ranged from 13 to 18.

In October 1990 during a set form period, every girl in the class was handed an envelope containing an explanatory letter and questionnaires. Each girl was asked to take the envelope home, to discuss it with her parents and to return the sealed envelope within 3 weeks to the school whether or not the family had decided to participate.

The enclosed letter explained the objective of the project, ie that norms were required for questionnaires being developed and used in a project on families and illness. It also explained that the questionnaires were anonymous and that the code numbers on the questionnaires were merely a way of keeping families linked together. A telephone number was supplied for any further questions. On a separate sheet families were asked for details about the composition of their households and whether any child in their household had been referred to a specialist in the past 2 years for certain specified illnesses.

All family members over the age of 13 currently living at home were asked to complete questionnaires. The FAM scale was designed for the 13 and over age group (Skinner, 1983). Step parents/cohabitees were included if they had been in the household for more than 3 months. The questionnaires were the Family Assessment Measure (Skinner, 1983)) and the piloted items of the Edinburgh Family Scale.

9.2.2 Response Rate

Out of the total pool of 104 pupils, 65% of the sample pupils completed the questionnaires. This ranged from a low 50% in one class to 75% in another. As the questionnaires were anonymous, it was difficult to establish the reasons for refusal. There were several points at which non-response could occur. The girls may not have taken the questionnaires home. Of those questionnaires that got home, parents may have refused to participate. Finally completed questionnaires may not have made their way back to school. The timing of the approach was in retrospect

unfortunate. It was just before a break for a long half term holiday when perhaps other preoccupations took priority. Doubt must remain over the representativeness of the sample.

9.3 Procedure School B

9.3.1 Subjects

A sample from a second school (school role 1025) was taken in February 1991. A second sample was needed both to continue the development of the Edinburgh Family Scale by further item analysis and factor analysis and to include a sample of boys, 50% of the cystic fibrosis group and 29% of the well family group in the main study being male. This school was a large mixed private secondary school with a slightly higher ratio of boys to girls at the school. Again no formal analysis of social background was made but the majority of pupils would be from social classes I, II and IIIa, with a higher proportion from IIIa than at School B.

This school organised its classes in 3 academic ability bands and in the upper part of the school in subject choice bands. 2 classes from each of the 5 school years were selected from the middle ability band with a balance of science and arts specialists in the higher forms. Age of pupils ranged from 13 to 18.

In order to improve the response rate following the rather low response rate at School A, the Head agreed to an opt out rather than an opt in procedure. All pupils were sent home with an explanatory letter about the project with a tear off slip to be returned if the family did not wish to participate. Only pupils returning such a slip were permitted to absent themselves from the pupil test sessions which took place in 2 supervised sessions with half the children at each. Every pupil attending completed the FAM scale and Minuchin test items. Each pupil was given an envelope to take home containing questionnaires, this time not for all family members over 13 but

for parents and step-parents living in the household only. Each envelope was ready stamped and addressed for direct mail return. Parents were asked to complete the FAM, the Minuchin items, details of household composition and children's illness in the past 2 years as at School A. In addition, they were asked to complete the modified Life Events Schedule in order to provide some norms for the main study (see below for details).

9.3.2 Response Rate

Efforts to improve response rate for the pupils were rewarded. Out of a total pool of 163 pupils, 134 completed the questionnaire. The pupil response rate ranged from 100% to 68% of each class. 4 families refused, 10 pupils were absent from school on the test day and the remaining 15 were mysteriously absent from the test centre (mostly from one class). However, only 60% mothers and 33% of fathers returned the questionnaires. As only 4 families had refused to enter the study, it seems highly likely that many questionnaires never reached the parents. The representativeness of the parental sample, particularly the fathers, must remain in doubt although it was possible to see if there were differences in the pupils between responding and non-responding parents.

9.4 Sample Characteristics

The sample characteristics of the schools A and B were very similar and are presented in Table 9.1. Average age of fathers was 46, of mothers 43.5, and of adolescent responders, 15. The median age of children in the family (the family stage) was 14.5 - so the prevailing family culture was that of young adolescent. 95% of families were 2 parent families. 5% per cent of these were reconstituted families. Approximately 80% of the families had either 2 or 3 children.

TABLE 9.1
CHARACTERISTICS OF SUBJECTS IN SCHOOL A AND SCHOOL B

	School A	School B
Sample Size	N	N
Index Pupil Pool	104	163
Fathers	42	44
Mothers	49	79
Daughters	65	58
Sons	15	74
Age	Mean (SD) Range	Mean (SD) Range
Fathers	46 (4.6) (39-60)	46 (5.2) (37-63)
Mothers	44 (4.3) (37-56)	43 (3.3) (37-50)
Daughters	15 (2.1) (12-23)	15 (1.3) (13-17)
Sons	16 (2.6) (13-21)	15 (1.4) (12-18)
Family Stage	Mean (SD) Range	Mean (SD) Range
Median Age	14.6 (3.25) (8-23)	14.6 (2.05) (10-19)
Family Size		
2 parent family	95%	94%
1 child	6%	10%
2 children	35%	48%
3 children	51%	31%
4 children	8%	10%
5 children	0%	2%

9.5 The Family Assessment Measure

9.5.1 Background

The Family Assessment Measure (FAM-III) (Skinner et al, 1983) is based on the process model of family functioning which was originally developed by Epstein and his colleagues (Epstein et al, 1983). This model states that family members share common goals and that to achieve these goals certain family tasks must be carried out in the context of important interpersonal processes. There are 3 different versions of the FAM: a general scale, a dyadic relationship scale and a self rating scale.

The most widely used scale is the general scale which measures family functioning as a whole. This is a 50 item scale breaking down into 7 subscales measuring the process model's critical family characteristics: task

accomplishment, role performance, communication, affective expression, affective involvement, control, and values and norms. These can be averaged to provide an overall score. In addition, there are 2 subscales measuring response bias: social desirability and denial. The scale's psychometric properties and applications are described in Skinner et al (1983), Steinhauer (1984) and Skinner (1987). The FAM-III Administration and Interpretation Guide provides normative data from a normal family sample for adults (n = 247) and adolescents (n = 65). These norms have been standardised to have a mean of 50 and a standard deviation of 10. The higher the score, the more dysfunctional the family. Skinner suggests a cut-off of 60 for a dysfunctional family but does not make it clear why a cut off of one standard deviation is chosen.

The discriminating power of FAM-III in differentiating problem from non-problem families was examined by Skinner et al (1983) but unfortunately little information on the problem families is provided. Limitations of the FAM-III centre on the limited reliability and validity studies completed to date. There are high correlations between subscales suggesting that the constructs within scales are not distinct. No test-retest reliability data is available. Published normative data is very limited, particularly for adolescents. Information is lacking on FAM scores at different stages in the family life cycle.

To date there are no published norms for use in a British population and it was with the objective of rectifying this that the scale formed part of the schools battery.

9.5.2 Sample Characteristics

Full details of the sample and test procedure are given above. The mean age of Skinner's normal sample of children was 14.9 years, closely resembling the mean of the Schools A and B sample. His adults were slightly younger (mean age 38) than in this sample. About 22% of his

sample had previous marriages, a significantly higher proportion than in this sample (5%). The social class characteristics of his sample are close to this sample.

9.5.3 Mean Scores

As there were no significant differences between schools, data has been pooled for Schools A and B and are presented separately for adults and adolescents in Tables 9.2 and 9.3. An examination of mean scores and the 95% confidence intervals of these means shows that adults' scores match closely the Canadian norms which were standardised to Mean 50 and SD 10. Skinner included the Denial and Social Desirability scales as a check on response bias. Mean scores of these scales for Scottish adolescents are low (Denial Adolescent Mean = 44.4 and Social Desirability = 48.0) and Denial is low for adults (Adult Mean = 48.1). This increases confidence that the other subscale means are a true reflection of attitudes. The adolescents have quite low scores on affective expression (Mean AE = 43.5). Other mean scores for adolescents are marginally raised. Skinner (1987) suggests that a difference of 10 points on a subscale signifies a clinically significant difference. None of the means approach such a large difference.

9.5.4 Reliability

Skinner (1983) gave internal consistency reliability estimates for the FAM-III subscales (Cronbach's alpha). These range for the seven subscales from .78 to .65 for adults and from .75 to .60 for adolescents. In the present sample, alpha co-efficients are a little lower (Adults range .91 to .51 and Adolescent range .91 to .56 See Tables 9.2 and 9.3 for details). It would be difficult to maintain high co-efficients across samples with only 5 items on each subscale. Nevertheless, anything below .60 (Kline, 1986) should be treated with caution.

9.5.5 Validity

Skinner (1987) examined the FAM general scale profiles of husbands and wives in the same family in normal couples and in what he describes as "clinical" couples. If the scale has any external validity, there ought to be some correspondence within families on their test profiles. He found a moderate level of agreement in normal families (Mean correlation r = .36, SD = .43) and slightly stronger agreement for the "clinical" families (Mean r = .51, SD = .41).

In the Scottish schools sample the association between family triad of father, mother and child was calculated. In interpreting these results it should be recalled that data was available for only 47% of fathers and 70% of mothers so that calculations must take place on a subset of data. In order to forestall Type 1 errors by calculating a bank of correlation coefficients, it was decided to examine the FAM Overall score (the mean of the 7 subscales) only. The correlation between child and mother was .51, between father and child .52 $p < .001$. The correlation between mother and father was .23 $p < .07$.

9.5.6 Responding v non-responding parents

Concern was expressed earlier that the rather low participation rate on the part of the parents might make the representativeness of the sample doubtful. At school B participation of pupils was very high so it was possible to compare the FAM scores of the children whose parents responded with those who did not. Non-responding parents might have more dysfunctional families for example. The difference between the child sample mean Overall FAM score in responders and non-responders was 2.33 score points, with a 95% confidence interval from 0.75 to 5.92; the t test statistic was –2.54, with 139 degrees of freedom and an associated p value of $p = 0.01$. Thus non-responding parents had children who saw their families as being slightly more dysfunctional than children with responding

parents. The difference, however, is clinically insignificant and an attempt to sort children into responders and non-responders on the basis of their FAM scores with a discriminant function analysis failed completely.

TABLE 9.2 FAM-III GENERAL SCALE
Mean Scores and Internal Consistency

Adults (n = 214)

Subscales	Mean	(SD)	95% Confidence Interval	Cron-bach's alpha
General Scale	50.4	(6.3)	49.4 to 51.3	.91
Task Accomplishment	48.1	(8.4)	46.9 to 49.2	.60
Role Performance	51.7	(9.4)	50.5 to 53.0	.70
Communication	51.0	(7.9)	49.8 to 52.0	.51
Affective Expression	50.5	(8.1)	49.3 to 51.6	.67
Affective Involvement	50.0	(7.9)	48.8 to 51.0	.75
Control	50.3	(8.3)	49.1 to 51.4	.65
Value and Norms	51.8	(7.8)	50.8 to 52.9	.59
Denial	48.0	(8.9)	46.7 to 49.2	.68
Social Desirability	50.4	(9.6)	50.0 to 52.6	.90

TABLE 9.3 FAM-III GENERAL SCALE
Mean Scores and Internal Consistency

Adolescents (n = 214)

Subscales	Mean	(SD)	95% Confidence Interval	Cron-bach's alpha
General Scale	51.3	(7.5)	50.1 to 52.5	.91
Task Accomplishment	52.4	(9.3)	51.1 to 53.8	.61
Role Performance	54.2	(9.2)	52.9 to 55.4	.56
Communication	53.0	(10.3)	51.6 to 54.4	.63
Affective Expression	43.5	(10.3)	42.1 to 45.0	.62
Affective Involvement	51.1	(9.7)	49.7 to 52.4	.71
Control	52.3	(7.8)	51.2 to 53.4	.61
Value and Norms	51.1	(8.8)	49.9 to 52.3	.63
Denial	44.4	(9.7)	43.0 to 45.7	.71
Social Desirability	48.0	(10.5)	46.5 to 49.4	.86

9.6 The Edinburgh Family Scale

9.6.1 Background

Minuchin measured his "psychosomatic" family dimensions by using therapist assessment scales to rate both a family task and a diagnostic interview. If a reliable and valid self report questionnaire gauging the same concepts could be developed, this would have the advantage of being less cumbersome to administer and score.

There have been several attempts to produce a self report inventory that would operationalise Minuchin's "psychosomatic" family dimensions: enmeshment, overprotection, rigidity and lack of conflict resolution. Burbeck (1979) developed the Index of Family Characteristics (IFC) as a tool to assess the relationship between severity of illness and the psychosomatic transactional dimensions in the families of 42 asthmatic

children. The questionnaire is composed of 4 subscales representing the 4 Minuchin psychosomatic dimensions. There are 4 items in each subscale. For example, the statement "When something happens to one person in my family, the other family members find out about it" is rated on a 6 point Likert scale ranging from "almost never" to "almost always". The content validity of these items is good, being clinically recognisable as derived from Minuchin's conceptualisation. The author states that the scale was developed exclusively for this research project. In the absence of any information to the contrary, one assumes that the scale has been tested only on the 42 patients in the study. Internal consistency measured by coefficient alphas are quoted but are low, not altogether surprising with only 4 items in each subscale. There is a brief mention of a factor analysis having confirmed the existence of 4 dimensions. No details are given but a sample size of 42 would make this a dubious endeavour. In conclusion, then, the item content looks promising and seems clinically appropriate but the psychometric properties of this scale remain underdeveloped.

After item selection by 6 family therapist judges and a pilot study on "fifty volunteer families", Perosa and colleagues (1981) developed a questionnaire (the Structural Family Interaction Scale) consisting of 13 primary subscales of 5 items each. The subscales are termed: enmeshment, disengagement, overprotection, neglect, rigidity, flexibility, conflict avoidance, conflict expression without resolution, conflict resolution, parent management, triangulation, parent child coalition and detouring. There are also 10 subscales of 2 items each. Responses to the 85 items are made on a 4-point scale of agreement ranging from "very true" to "very false". 2 adults and 2 children (mean age 12) from 50 further families, half of whom had a learning disabled child, completed the questionnaires and 6 of the scales were able to differentiate the learning disabled from the non-handicapped families (Perosa and Perosa, 1982). Alpha co-efficients on the scales varied from .71 to .93. Perosa and Perosa (1990) state that the SFIS "has differentiated family variables associated with clinical populations from those related to non-clinical control groups" but no details are given. It is

not possible to provide a comprehensive assessment of the SFIS given its current state of development and the lack of published data which has not been made available through personal communication.

A larger scale study has been carried out by Kog at the University of Leuven (1985). The families of 50 eating disorder in-patients and a representative sample of 220 non-patient control families completed in its original form a 106 item scale. Statements such as "Conflicts do not occur in our family" have 6 response possibilities ranging from "definitely not true" to "true all of the time". The items described "the various components that Minuchin and authors associated with the 4 psychosomatic family features". From a factor analysis (Varimax rotation) 3 scales are arrived at: conflict (30 items), cohesion (21 items) and disorganisation (22 items). These form the "Leuven Family Questionnaire". An examination of the "cohesion" items suggests that the sharing and closeness aspects of Minuchin's enmeshment and overprotection concepts are covered but the "intrusive mind reading" aspects are not. "Conflict" seems to cover conflict behaviour generally but no items seem to reflect the conflict avoidance/diffusion aspect of Minuchin's concept. The final subscale Kog describes as "disorganisation" which Kog claims to measure poor intergenerational boundaries (one aspect of enmeshment). Unfortunately the items bear little resemblance to the concept but seem instead to refer to a generally negative family evaluation. No information is supplied on item-total correlations but as the scale is long, internal consistency is high. The scale was able to discriminate between clinical and non-clinical families on "disorganisation" and "conflict" but not on "cohesion". The author says that she was unable to operationalise Minuchin's concept of "rigidity". It may well be that items tapping Minuchin's concepts did not survive item analysis but as it stands the scale is not fully recognisable as being a measure of "the psychosomatic family".

9.6.2 Initial item pool

Development of the scale was closely modelled on procedures suggested by Nunnally (1978) and Kline (1986).

A pool of 94 items was formed from ideas generated by Minuchin's clinical description (Minuchin, 1975), from previous attempts at a scale (eg Burbeck, 1979 and Kog, 1985) and from suggestions from an experienced family therapist. These items were submitted to 4 family therapists (minimum clinical experience 6 years) who were given Minuchin's own descriptions (1975) of the 4 family transactional dimensions of "enmeshment", "overprotection", "rigidity" and "lack of conflict resolution" and asked to select the 9 or 10 items that best captured the flavour of the 4 concepts. Items endorsed by at least 3 of the 4 therapists were shortlisted. These items were piloted on 5 families leading to a further rejection of items due to ambiguity, difficulty with double negatives and lack of comprehension. A final list of 48 items, 12 for each subscale, were chosen for inclusion in the schools test battery.

9.6.3 Test format

Nunnally (1978) has demonstrated that the reliability of a Likert scale increases with the number of scale steps and that this decreases sharply after 7 scale steps. A 7 point scale would have been the format of choice. However, a significant constraint on the study was that the whole testing procedure had to be completed within one 40 minute school period. In the interests of simplicity and speed, it was decided to follow a format identical to the 4-point scale Family Assessment Measure (Skinner, 1983) which was also being administered. The format was as follows. Each respondent was instructed that:

> "On the following pages you will find some statements about your family as a whole. Please read each statement and decide how well the statement describes your family. Put a tick in the box that best describes your family."

Statements were rated on a 4 point continuum "strongly agree", "agree", "disagree" and "strongly disagree".

9.6.4 Sampling methods and test procedure

Full details of the sample and administrative procedure are given in the general description of the schools study (see above). Kline (1986) suggests that for a satisfactory item analysis either 1 large representative sample or 2 smaller ones, minimum size of 100 each, is needed. This criterion was met by 171 subjects from School A and 257 from School B completing the pilot questionnaire.

9.6.5 Item analysis

Item analysis was carried out using the item analysis module of the statistical package CSS: STATISTICA.

Analyses were performed separately for each school and for adult and adolescent groups. Preliminary examination of the correlation matrix for both schools suggested that the enmeshment and overprotection subscales were highly correlated and clearly measuring the same dimension. One of the first decisions made was to collapse these 2 subscales for all further analysis.

Item-total correlations (Pearson product moment with pairwise deletion of missing data) were calculated for the 3 reconstituted subscales. At this stage clear outliers were rejected as were, in the interests of increasing variance, items that were endorsed by more than 85% of the sample in one direction. Items which were satisfactory for the adolescent group but not

for the adult group and vice versa were also rejected so that the same questionnaire could be used for both populations.

At this point a principal components analysis (orthogonal Varimax rotation) was carried out on the School A sample. The Cattell scree test (Cattell, 1966) suggested a 3 or 4 factor solution for the data. A 3 factor solution proved the most satisfactory because item loading on these factors matched closely to the hypothesised 3 Minuchin dimensions of "conflict", "rigidity" and "overprotection/enmeshment".

The final selection of items for inclusion in the Minuchin scale was made using both statistical and clinical criteria. Nunnally (1978) suggests that factor loadings of each item should be greater than .3 and should load more highly with one factor than with the others. This cut off left the item pool with 11 "good" items for the conflict scale, 10 "good" items for the enmeshment/ overprotection scale and 8 "good" items for the "rigidity" scale. 3 further items were rejected for being too similar in content to other items. Internal consistency already seemed high enough to justify this step. One item which did not quite meet the .3 factor loading criterion but was considered clinically important was reinstated on the rigidity scale. No item correlated more highly with another subscale total than with its own.

9.6.6 Replication

A further factor analysis of the remaining 27 item scale was carried out on the School B sample (n = 257) to test the stability of the dimensions. The questionnaire stood up to this test well. Although not surprisingly there were differences in the strength of factor loadings for some items, there were no material changes of an individual item now loading more highly on a different factor. The factor loadings for the pooled scores of both schools on the 27 item scale are presented in Table 9.4.

TABLE 9.4 EDINBURGH FAMILY SCALE: FACTOR LOADINGS

School and School B combined n=323	Raw Varimax Rotation Normalised Factor Loadings (3 factor solution)		
	Factor1	Factor2	Factor3
Enmeshment/Overprotection			
When somebody in our house gets hurt or upset, we all react	.58	-.13	.10
We prefer doing things at home to going out on our own	.37	.24	-.13
I feel responsible for family members	.57	-.07	-.11
We are very concerned about each other	.73	.02	-.19
Parents are always around for the children	.47	.17	-.14
My parents/children are just like close friends	.40	.23	-.23
Family ties are more important to us than friendships	.55	.23	-.09
When one family member has a problem, everyone worries about him or her	.71	-.04	-.12
Family members are very involved in each other's lives	.66	.14	-.07
Rigidity			
We like to smooth things over	.23	.39	.14
Other people's suggestions about our family tend to be rather a waste of time	-.09	.35	.13
We see no need to change our way of doing things	-.01	.61	-.43
Old ways of doing things tend to be the best	-.05	.54	.07
It's better to go along with what other people say in the family	-.03	.50	.20
We prefer things to stay the same in our family	.06	.73	-.08
Our way of life does not need to change	.06	.60	-.24
In our family we like things to be cut and dried	.10	.46	.16
Rules change in our family	.14	.22	-.20
Conflict			
We spend too much time arguing about what our problems are	-.05	-.04	.59
When problems come up, we try different ways of solving them	-.28	-.03	.39
We never let things pile up until they are more than we can handle	-.11	-.13	.45
When things aren't going well, it takes too long to work them out	-.15	-.12	.60
We deal with our problems even when they are serious	-.29	.09	.44
We have disagreements that can't be talked about	-.24	.04	.41
The least thing can cause an upset in our family	-.07	-.09	.58
We never seem to get to the bottom of family problems	-.20	.00	.73
The same old arguments come up again and again	-.01	-.09	.73

9.6.7 Internal consistency

Co-efficients of reliability (Cronbach's alpha) were calculated for each subscale. These are presented separately for adult and adolescent groups in Table 9.5 and range from .60 to .76. Kline (1986) suggests that alpha should not fall below .60 for an individual scale and this criterion has been met.

TABLE 9.5 EDINBURGH FAMILY SCALE: INTERNAL CONSISTENCY RELIABILITY ESTIMATES AND AVERAGE INTER-ITEM CORRELATIONS SCHOOLS A AND B COMBINED

Subscale	Adult (n = 189)		Adolescent (n = 192)	
	alpha[1]	r (item)[2]	alpha	r (item)
Enmeshment	.74	.25	.74	.25
Conflict	.76	.27	.76	.27
Rigidity	.73	.23	.60	.13

[1] Cronbach's alpha co-efficient
[2] Average inter-item correlation

9.6.8 Scoring

Each item is scored on a scale of 0 to 3 and the items for each subscale are summed to form raw score totals. There were differences between the mean raw score totals of the 3 subscales and differences in the distribution of these scores. There also were significant differences between the mean scores for the adults and adolescent population on the enmeshment and conflict subscales (see Table 9.6). The only difference between schools was a clinically insignificant difference of .5 on the Enmeshment subscale (see Table 9.7), a difference accounted for by a slight tendency for women to score more highly on enmeshment than men. Therefore results for schools were pooled. It was decided that norms could be expressed more satisfactorily with standard scores using a Z score transformation, using the common convention of a distribution with a mean of 50 and a standard

deviation of 10 (Kline, 1986). Transformation tables have been produced separately for adult and adolescent populations. The final scale and the transformation tables are reproduced in Appendix V.

TABLE 9.6 EDINBURGH FAMILY SCALE: RAW SCORE TOTALS SCHOOLS A AND B COMBINED

Subscale	Adults	Adolescents		
	n Mean (SD)	n Mean (SD)	t	p value
Rigidity	190 13.3 (2.9)	176 13.4 (3.0)	0.50	.61
Enmeshment	191 17.3 (2.8)	183 15.9 (3.4)	6.97	**<.0001**
Conflict	199 9.5 (2.9)	192 11.4 (3.7)	5.73	**<.0001**

TABLE 9.7 EDINBURGH FAMILY SCALE RAW SCORE TOTALS X SCHOOLS

	School A	School B		
Subscale	n Mean (SD)	n Mean (SD)	t	p value
Rigidity	141 13.6 (2.9)	225 13.1 (2.9)	1.45	.14
Enmeshment	144 16.6 (3.0)	230 15.9 (3.4)	2.08	**.04**
Conflict	155 10.0 (3.3)	236 10.7 (3.6)	1.93	**.05**

9.6.9 Discussion

How well did Minuchin's concepts stand up to an item reduction from the 48 items administered to the school population to the final 27-item scale? The enmeshment/ overprotection scale retains the closeness, cohesion, over-protective and poor generational boundary aspects of Minuchin's dimensions. What has been lost are items that tap Minuchin's "mind reading concept", for example "we know what other people in the family are thinking". Such items are also absent from the Leuven Family Questionnaire (Kog, 1985). What one is left with is a concept that from its item content appears to be very similar to the "cohesiveness" dimension that

studies of other measures of family function elicit. Convergent validity studies find that the Cohesion scale of the Family Environment Scale (Moos and Moos, 1981), the Affective Involvement Subscale of the Family Assessment Device (Epstein, Baldwin and Bishop, 1983) the Cohesion scale on FACES III (Olson, Portner and Lavee, 1985) all seem to be self ratings of global family closeness (Dickerson and Coyne, 1987; Bloom, 1985; Perosa and Perosa, 1987). Families evaluate "cohesion" as a positive family attribute and it is viewed as a functional attribute in the Family Scales cited above. However, clinicians see "enmeshment/overprotection" as a negative attribute. The concept includes the idea of cohesiveness but is not defined by it.

The "lack of conflict resolution" concept has stood up rather well. The retained questions tap both the idea of conflict being present and the notion that conflicts tend not to be out in the open.

On the rigidity scale, items that were lost were ones that had a high social desirability content leading to almost universal endorsement of the item and therefore minimal variability.

What was the relationship between the 3 subscales? Minuchin would predict that there would be small correlations between the scales. There was a positive correlation between the overprotection/enmeshment scale and the rigidity scale ($r = .22$) However, the conflict scale correlated negatively with both these scales ($r = -.43$ and $r = -.29$). In other words, in the normal population, families who saw themselves as having no difficulty in resolving conflict also saw themselves as close and cohesive and disliking change. This was subsequently found to be true for the clinical population studies in the main project. As it stands, this runs counter to Minuchin's concept of the "psychosomatic family" in which the enmeshed family also has difficulty in dealing with conflict.

9.7 The Life Events Scale

9.7.1 Background

Families may appear dysfunctional or under stress because of factors related to their child's illness. However, it is at least plausible that family stress is related to stressful life events independent of the illness factor. Therefore it was decided to include a measure of life events in the main study.

There are two main methodologies for collecting data about life events: the checklist approach and the panel approach. The checklist approach was developed by Holmes and Rahe (1967) with the Schedule of Recent Events (SRE) and from this there have been numerous derivative instruments (eg Paykel et al, 1971; Dohrenwend et al, 1978). In the checklist approach subjects are given a standard list of potentially stressful events with predetermined weighted scores. Scores are then are summed to provide an index of summated life stress for the time interval under investigation.

The panel approach was developed by Brown and Harris (1978) and the best known example of this is their London Life Events and Difficulty Schedule (LEDS). The LEDS is a lengthy semi-structured interview for eliciting information on life events and the subject's social background and makes use of an extensive glossary of definitive and elaborate scoring rules. The amount of stress involved for the individual under study is determined, after all the relevant information has been gathered by an interviewer, by a panel of raters whose judgments are pooled to form a consensus rating on a 4 point threat scale.

The advantage of the checklist approach is that it is quick to administer and score and does not require an elaborate superstructure of trained interviewers and raters. However, Brown (1981) and others (eg Katschnig 1986) have criticised and rejected the checklist methodology.

The main plank of this criticism has to do with reliability issues. Katschnig (1986) found that at the level of the individual patient there was virtually no agreement between stressful events elicited by the LEDS methodology and those elicited by the Holmes and Rahe's checklist. He also found low 2 week test-retest reliability on the SRE. However, there may also be reliability problems with the LEDS methodology. Although satisfactory reliability co-efficients have been obtained between raters for "level of threat" on vignettes of events given to those raters (eg Parry et al, 1981), information is strangely lacking on whether the events themselves can be reliably elicited from subjects either by test-retest (same interviewer) or by test-retest (using two interviewers). Hudgens and colleagues (1970) found only 57% agreement between the accounts of patients and additional respondents about events occurring in the previous 12 months which is not very encouraging.

Some of Brown's (1978) criticisms apply to the Holmes and Rahe instrument in particular rather than to the method in general. Vague items can be rewritten to make them clearer, reliability can be improved by making event categories more precise and methods of analysing results need not assume additivity of qualitatively different types of events.

A criticism of the Holmes and Rahe methodology is that pre-determined weights of stress events take no account of the context of that event for individuals, a factor which the LEDS methodology is able to do very well. A simple solution to this problem would be to ask subjects to give their own weighting to the life events they have experienced. The argument against this has been to suggest that subjective rating would confound the relationship between stressful events and psychopathology. However, as Brown and Harris (1978) (p.114) themselves report high levels of agreement between objective consensus rating and the self report of threat (95% for the general population and 84% for depressed patients), taking account of chance associations, it is hard to see how this objection can be sustained.

As criticisms of the checklist approach did not seem sufficiently powerful to cancel out the clear advantages of this approach in terms of cost and speed, a new life events check list was developed for use in the families and illness project, salient for families with adolescents and taking into account some of the criticisms of the checklist approach. This checklist was administered as part of the schools battery.

9.7.2 Procedure

A list of 22 events modified from Miller and Salter (1984) was drawn up designed to cover events that would be salient to British households with adolescents and young adults living in them. Instructions were that the schedule was to be completed by one parent with the help of the other parent if wished. The respondent was asked:

> Has this happened to anyone in your family circle in the past six months?

and asked to tick Yes or No. "Family circle" was defined as anyone living in the household or any relative that the respondent was very close to. Parents were asked not to score the same event twice, eg both major change in quality of marital relationship and divorce or break up of special relationship.

If subjects answered 'Yes' to a particular event having happened in the past 6 months, they were then asked to estimate how much of a problem the event had been to their family. This was on a 5 point scale ranging from 0 = no problem to 4 = a major problem. It was thus possible to score the scale in two ways. Firstly by simply summing the number of events and secondly by summing the problem weights.

9.7.3 Incidence of Events

84 parents from School B completed the life events schedule. The incidence of individual events are presented in Table 9.8. Also presented is the incidence of those families who described those events as being of "some problem" through to a "major problem", ie a rating of 2, 3 or 4 on the 5-point scale and also of those events that were a "major problem" (score 4), equivalent to "severe threat" on the LEDS. Raw scores are presented rather than percentages to avoid distortion of event/severity ratio.

Sitting important exams is the most common life event reported by these families. Median age of children in the family was 15 so this is not altogether surprising. What is of note is that over 50% of the families reported that exams had been a family problem. Trouble with relatives outside the home, theft and damage to property and major changes at work were both common and stressful events.

The proportion of families having experienced one or more events that were major problems (severe threats) was 14%. This is much in line with the median score of 17% for control groups that Brown found in his review of studies of life events (Brown and Harris, 1986).

9.7.4 Life Events and Illness

Parents were asked if any of their children had been referred to a specialist or had had hospital treatment for any of a specified list of illnesses in the past 2 years. Rates for individual illnesses were too low for separate analysis so the data was dichotomised into presence or absence of referrals. No correlation was found between presence of illness and life events measured either by number of events or by problem weights.

This data should provide useful data from which to evaluate the significance of life events data in the main study.

TABLE 9.8 LIFE CHANGE EVENTS
INCIDENCE OF EVENTS AND EVENT PROBLEMS IN 84 SCHOOL B FAMILIES

Has this happened to anyone in your family circle in the past six months?	Incidence	Problem	Major Problem
1. Retired from work	1	1	1
2. Started or left a job	21	7	1
3. Started or left school or college	20	4	0
4. Had trouble at work	9	5	0
5. Had a major change in work conditions or responsibility	18	9	1
6. Sat important exams	34	17	0
7. Got engaged or married	1	0	0
8. Family member left home	8	3	1
9. Became pregnant	0	0	0
10. Had a baby	0	0	0
11. New person joined the house	2	1	0
12. Had trouble with relatives (outside household)	18	11	4
13. Death of person in household	0	0	0
14. Death of other close family member or friend	11	8	3
15. Major accident or injury	1	1	1
16. Major change in health of family member (other than child in research study)	6	4	3
17. Major change in quality of marital relationship	4	3	3
18. Divorce or break up of special relationship	6	6	3
19. Major change in financial situation	9	7	2
20. Loss, damage or theft of property	19	11	2
21. Had trouble with the police or law	1	1	1
22. Moved house or change in housing conditions	13	7	3

CHAPTER 10: DISCUSSION

10.1. Methodological Considerations

10.1.1. The Sample

The generalisability of research findings depends to a large extent on the representativeness of the sample and the adequacy of the control groups. The AN sample was typical of the client group referred to secondary and tertiary treatment resources who form the bulk of the research population but may not be characteristic of community populations such as those in the Swedish study (Rastam & Gillberg, 1991).

Difficulty in recruiting sufficient AN patients in the time-scale led to 10 patients being included in the study who no longer met the weight criterion for AN. This meant that a few of the families felt that their circumstances had improved considerably and in fact said that they would have given much more extreme scores a few months back on, for instance, the Impact on Families scale.

The CF families proved for the most part a satisfactory comparison group. Although they had had considerably longer to adapt to illness than the AN group the fact that some of the young people were in the final stages of their illness and that others had had recent crises such as first hospital admission meant that family burden was comparable with the AN group. Furthermore median duration of illness in the AN group was 3 years so that this group had had the opportunity to make some kind of adaptation. This was reflected in the very similar Impact on Family scores in the 2 groups.

The well families were if anything "superwell" by virtue of being nominated as healthy by the ill families or by being selected from GP lists. 30% of families on the GP lists were rejected through failing to meet the health criterion.

Subjects who refuse to take part in research may not be an unbiased sample. There was anecdotal evidence that some of the refusers in the two ill groups did have family problems. However, there was no reason to suspect that family dysfunction was more likely in CF or AN refusers so sample bias is unlikely to explain differences between those groups. The refusal rate in the well group was very low .

A shortcoming of the samples was that the CF group was not matched with the AN and well groups for sex and social class. In fact this made, perhaps surprisingly, little difference to the findings. Lower social class subjects and female subjects were slightly more likely to describe themselves as Enmeshed than others subjects. Clinically teen-age boys were observed to be more monosyllabic on the problem solving task than teenage girls although this was not true of the young adult men. However this did not effect their ratings.

Although the initial power analysis indicated that sample size was adequate to detect clinically significant differences between groups, for certain purposes it was too small. It was insufficient for an adequate test of life events issues(see 10.3). It was too small to detect relationships of interest in the follow up study: the differences between the rather small correlations typically found in most psychiatric research. This was not due to high attrition between baseline and follow-up which was in fact less than most follow up studies. In order for the correlation between changes in overprotection\enmeshment and changes in weight for AN ($r = .40$) and CF ($r = .18$) to have been significantly different at the .05 level, sample sizes of 140 in each group would have been needed.

10.1.2. Baseline to Follow Up

The time between baseline and follow-up was 8 months. In retrospect this was too short to allow for much significant change to take place in health status in either of the ill groups making it difficult to assess the relationship between health status and family functioning over time. A

more satisfactory design would have had a gap of perhaps 2 years though it is likely that this would have meant greater attrition of the sample.

10.1.3.Measurement Issues

A strength of the study was the decision to use self report, interview and observational methodology to measure family interaction. This had also been the approach of the Belgian study (Kog et al, 1989). Most published research on eating disorders and families relies heavily on paper and pencil tests, probably because they are convenient to use and easy to score. The differences between groups in this study were not picked up by the self report measures which illustrates the dangers of the unitary approach.

The Expressed Emotion Index is well established as a robust measure of negative affective style and intrusive overconcern. As scoring the taped interviews involved subjective decisions on the part of the rater, a design limitation was that rating was carried out by the author who was not blind to the objectives of the study. However, this weakness was in part offset by a sample of tapes also being rated by an experienced independent assessor. This resulted in good inter-rater agreement and no evidence of systematic bias.

The Problem Solving Task had several strengths: it was conceptually similar to the methodology used in Minuchin's original work and the problems under discussion were salient to the household. A scoring system was devised that was modelled as closely as was feasible on Minuchin's own but hopefully removing some of the ambiguities of that system. A limitation of the design was that as with the EE index, ratings were carried out by the author who was not blind to the subjects or the objectives of the study. Again, this limitation was mitigated by a sample of interviews being rated by an independent assessor using audiotapes, transcripts and a scoring manual. Inter-rater reliability was good suggesting that bias may have been avoided.

The Morgan and Russell scale was administered by an independent assessor at baseline. At follow this was not possible. Again the possibility of bias was checked by an inter-rater reliability exercise.

10.2. Cystic Fibrosis Findings

10.2.1. The CF Patient

The most striking observation in the CF families was the robust psychological health of the CF patient. No patient reached DSM-III-R criteria for major depressive disorder and only 4 out of 29 were defined as "emotionally distressed" on the GHQ. Only 4 of the group had had a previous encounter with a GP or hospital for mental health problems. While personality was not formally measured the clinical impression was that these young people were indistinguishable from the well sample and quite distinct from the more troubled AN subjects. With increased age emotional distress was likely to be higher (although still low), an effect not found in the AN and well samples and not accounted for by severity of illness. These findings confirm the most recent research findings on CF adolescents and young adults (reviewed in Section 3.4.2) and mark a change from the dismal picture of the psychological functioning of CF patients in the early uncontrolled studies. They reflect the real improvement in quality of life now experienced by CF patients and the increase in longevity.

10.2.2. Mothers and Fathers

The finding that mothers of young CF children suffer raised levels of emotional distress, particularly depression, is well established. This is generally true for mothers caring for chronically ill children. The present study confirmed 2 uncontrolled studies (Boyle et al, 1976; Bywater, 1981) that this is also found in mothers of adolescent and adult CF patients and underlines the need for support for a minority of CF mothers to be continued well after the patient has reached the age of majority. The finding that fathers' mental health status was indistinguishable from well

group fathers again supported previous work in the field (reviewed in Section 3.9.2).

Results from the Camberwell Family Interview suggested that 31% of CF mothers were overinvolved (Expressed Emotion EOI) with their CF child. This compared with 19% of mothers of well children. Overinvolved mothers also tended to be those with raised levels of emotional distress. The early anecdotal accounts of CF adults and adolescents reported a high level of overprotection and infantalisation in mothers and argued that this was one of the main contributions to psychological malfunction in the CF young adult. In this study EOI in CF mothers was quite independent of the psychological health status of their child but was related to severity of illness. Perhaps the EE term "overinvolvement" has inappropriate negative connotations in evaluating the families of the chronic sick.

10.2.3. Family Interaction

The CF Families in this study were found to be functioning at a healthy adaptive level. Scores on the self report FAM measure were within the normal range confirming the finding of the Toronto study (Cowen et al, 1986) which had used the same instrument on a sample of CF adolescents and young adults and their families. They also confirm the findings of Shepherd et al (1990).

Observed interactions of CF families suggested that most families had low levels of criticism, good problem solving, with clear communication, ability to compromise and flexibility in problem solving strategies. Families were no more likely to show enmeshed overprotective behaviour than were the well families observed and less likely than the AN families. There were clearer boundaries between generations than in the other groups. These findings support Bywater's interview study in which she found CF teenagers to have good relationships with parents. In Chapter 8 an explanation for the success of these families was offered in terms of both

the collaborative alliance formed with their health workers over the years and the psychological well-being of their CF child.

The early anecdotal accounts of family functioning in CF patients which gave a more pessimistic picture of family life were largely carried out on the families of younger children and date from a time when the quality of life for CF sufferers was considerably worse than it is today.

There was no relationship between the family's own evaluation of its functioning and the severity of illness in the patient. Nor was there a relationship between observed criticism and problem solving skills and illness severity. Overprotection and enmeshed behaviour was more likely to be observed in patients who were low in weight although when lung function was added to the health index this effect disappeared. The large sample size in Cowen's study (n = 229) of CF families enabled very weak correlations between CF lung function and family functioning to reach significance whereas correlations of the same order in this study (n = 29) did not.

Changes in health status over the 9 month follow-up period did not result in change in family functioning. This had also been the finding in a 15 month follow-up of young CF children (Patterson et al, 1990).

10.3. Life Events and Family Functioning

It had seemed plausible that a life event deemed to be a severe threat to a family could have an impact on family functioning which might mediate any relationship between illness and family functioning. However, the presence or absence of threatening events did not appear to influence FAM scores, EFS scale scores or Expressed Emotion scores. Rates of severe life events in both ill groups were similar to those found in the Schools study and to those found in community studies (Katschnig, 1986).

These negative findings might be explained by design limitations. Most satisfactory life events research is carried out on samples of several hundred subjects. This is to accommodate the expected low incidence of severe threat. With a total sample of 87 families in this study, only 12 could be expected to experience severe life events, no more than 4 families in each group. Thus the sample was really too small to make any confident statements about the significance of life events. On the other hand the findings are congruent with previous research on CF patients and life events (Frydman,1981; Smith et al, 1983; Steinhausen, 1983) and AN patients and life events (Sohlberg et al, 1990).

10.4. Are AN Families More Dysfunctional than CF Families or Well Families?

10.4.1. Self Report

Results on the Family Assessment Measure suggested that AN families did not in general describe themselves as being more dysfunctional than either the CF families or the well families. Only 2 studies known to the author (Garfinkel et al, 1983; Garner et al, 1985) have used the FAM as a measure of family functioning in AN families. The AN patients in Garfinkel and colleagues' study reported more problems than well controls on 2 out of 7 FAM subscales and AN mothers were more dysfunctional on 4 out of 7 subscales. Their finding of no difference for fathers was replicated in the present study. A possible explanation for the slightly higher levels of dysfunction found by Garfinkel is that his AN sample consisted largely of bulimic-anorexic patients whereas only 4 out of the 27 AN subjects in the present study were bulimic. The literature reviewed in Chapter 2 suggested that patients with bulimia reported more family deviance than those without. Garner et al's study (1985) found that restricting anorexic families had FAM scores indistinguishable from test norms, but that the anorexic-bulimic and bulimic families had more dysfunctional scores, which would lend support to this explanation.

The literature looking at other self report studies of AN family functioning was reviewed in section 2.10.1. The negative findings here are not at odds with the conclusion of that review which was that AN patients and their parents did not see themselves as being any more or less cohesive or adaptable than control families. Raised levels of reported conflict were not found here which was the case in 50% of the studies reviewed. The remaining 50% of studies described raised levels of conflict.

Two recent studies (Wonderlich and Swift, 1990; Heinmaa et al, 1992) found that depressed mood in eating disorder patients mediated their evaluation of family functioning. In this study, emotional distress (GHQ), which includes depressed mood, was also found to correlate with self reported family dysfunction by the index child. Wonderlich and Swift suggest two possible mechanisms. Firstly, subjects with a history of hostile parental relationships may be particularly prone to develop depressed mood states. Secondly, the simple state effect of depressed mood may result in a state dependent and distorted perception of increased hostility in parental relationships.

Negative self evaluation by the household (Factor I), a composite score derived from all household members rather than the index child alone, was found to be independent of the index child's level of emotional distress, a finding that supports the second explanation rather than the first.

Also supporting the second explanation is the report from Lewinsohn and Rosenbaum (1987) who found that remitted depressives did not display the negative parental perceptions characteristic of persons in a depressed state. However the present study failed to find a relationship between improvement in the child's level of emotional distress and improvement in child's self report of family functioning over the 8 months follow-up period. The follow up phase of this project did have design limitations which will be discussed below.

Maternal emotional distress (GHQ) mediated negative self evaluation of household functioning, a finding well established in other research on families and illness (see Section 1.4.1).

Are there other possible explanations for the findings? The FAM was selected for inclusion in the assessment battery for 3 reasons: (1) it has reasonable psychometric properties (2) the items were less banal or inappropriate than other inventories investigated (3) it had been used on both the AN and CF populations in previous research. Nevertheless, many families disliked completing it, complaining that the questions were ambiguous or trite or unsuitable for their family stage. Therefore, doubts remain as to whether the scale is able to tap family functioning in a meaningful way. A second limitation is that the scale is a self report measure only. In clinical work, therapists search for meaningful explanations of behaviour of which the individual may not necessarily be fully aware (Eisler, 1988). Clinicians, or indeed any outside observer, may not see families as they see themselves.

10.4.2 Expressed Emotion and Families

The levels of Expressed Emotion found in the AN families were comparable to those found in previous studies of eating disorder patients (van Furth, 1991; le Grange, 1992a, 1992b). Szmukler et al (1985) reported EE levels that were slightly higher than those in this study but his sample included a wider range of eating disordered patients than van Furth and le Grange.

This study is the first known to the author in which levels of Expressed Emotion in AN families have been compared with the families of patients suffering from a non-psychiatric illness (Cystic Fibrosis). It is also the first to use the traditional Camberwell Family Interview methodology to investigate EE levels in the families of well adolescents and young adults. Florin et al (1992) recently examined the EE status of well controls in their

study of depression but they used the Five Minute Speech Sample methodology. The limitations of the FMSS were reviewed in Section 4.8.

Levels of Expressed Emotion were found to be higher in the households of AN and CF families than in well families. This was accounted for by higher levels of emotional overinvolvement rather than higher levels of criticism. Levels of critical comments in the AN parents were considerably lower than those found in studies of schizophrenia families (see Chapter 4 for review) and were no different from well families or CF families. Highly critical parents tended also to gain dysfunctional scores on the Family Assessment Measure and to show a high level of conflict on the problem solving task. They had no difficulty in disclosing their perspective on family problems but had few remedies for its resolution and seemed unable to compromise with other family members. This characteristic of inflexibility in high EE families has been well described by Vaughn (1986).

The finding that EOI was present more frequently in AN and CF families than in well families suggests that overinvolvement could be an illness related phenomenon, an hypothesis that gains further support from the finding that severity of illness correlated with high emotional overinvolvement. Some schizophrenia studies have suggested that overinvolvement may develop as the disorder becomes more chronic (Miklowitz et al, 1983; MacMillan et al, 1986) but in this study there was no relationship between duration of illness and overinvolvement.

Who were the high EOI families? As well as being more likely to have severely ill children in the household, the parents were more likely to show general psychological distress as measured by the GHQ. The association between high GHQ and high EOI was also found in the well households. In this study personality of parents was not measured directly. Van Furth (1991) has reported preliminary findings on the relationship between EE and personality and found EOI to be related to neuroticism. It could be that high EOI is a marker of neuroticism in parents. Further research in

the area of personality, mental state of parents and EE would be of great interest.

The finding that on the principal components analysis of household climate variables Factor II was composed largely of loadings on EE overinvolvement and overprotection\enmeshment observed on the problem solving interaction and that Factor III loaded largely on EE critical comments and observed problem solving skills gives some credence to the view that EE is indeed a measure of family interaction (Kavanagh, 1992).

10.4.3 Problem Solving Ability

Although the difference between groups was small, AN families were observed to be less effective problem solvers than well and CF families. An examination of the transcripts showed that this was not because they tended to brush problems under the carpet or that they were highly negative and critical but because once the problem was disclosed, AN families were unable to move on to the resolution phase: few positive suggestions were made, few compromises proposed. By contrast the CF and groups were quick to move forward, to suggest, to be flexible even if eventually they had to call a truce rather than find a solution.

10.4.4. Conclusion

There seems now to be some consensus from research that AN families do not show a highly critical affective style whether measured by observation (Goldstein et al 1981; Kog et al, 1989), EE criticism (Szmukler et al 1985; le Grange et al 1992a, 1992b; van Furth 1991) or self report (see 2.10.1. for extensive review). There is however a reluctance among researchers to embrace the most economical explanation for this which is that the families might be healthy, the preferred interpretation being that the families must be conflict avoiders. This was the position adopted by Minuchin. This proposition will be examined in the following section.

10.5. The Psychosomatic Family

Do AN families function as Minuchin's "psychosomatic" families: enmeshed, overprotective, rigid and low in toleration of conflict? The prediction was that AN families should be more "psychosomatic" than the CF families who in turn would be more "psychosomatic" than the well families.

10.5.1. Resolution of Conflict

Minuchin's concept of "lack of conflict resolution" is not a unitary one because it embraces 2 rather different behavioural styles. The first style involves denial of the existence of a problem or suppression and diffusion of it. He found that "some anorectic families agreed on everything instantly, denied or suppressed any mention of conflict. The second style involves "going on and on about poorly defined arguments without the problem conflict being brought to salience ... anorectic families went on and on about poorly defined arguments which they then rejected as unsuitable for the task" (1978). Minuchin's 2 styles are both poor problem solving strategies.

In this study AN families did not describe themselves as having more difficulties in resolving conflict (EFS Conflict scale) but they were observed on the problem solving task to be poorer problem solvers than CF or well families. Their strategies as described above (section 10.1.3) were characterised by lack of positive suggestions, lack of flexibility, and little risk taking rather than a high level of open conflict or negative affect. These characteristics are congruent with Minuchin's second "psychosomatic family" problem solving style.

Factor III on the principal components analysis loaded on EE criticism and on poor problem solving skills. Dare (1991) suggested a link between the EE critical comments index and Minuchin's concept of lack of resolution of conflict. However an examination of the transcripts showed that high Factor III families were poor problem solvers because they were high users

of open criticism, negative solutions justification and criticism strategies which are not prominent features of either of Minuchin's 2 psychosomatic problem solving styles.

In section 10.2.4. it was stated that there was now some consensus among researchers that AN families do not have a highly critical negative affective style. This has been shown in self report, EE criticism and observational studies. Researchers have been reluctant to accept the most obvious explanation which is that AN families are healthy. Instead they have invoked Minuchin's first problem solving style by arguing that families must be "conflict avoidant". The present study did not support this explanation. In this study the AN families were not observed to be more conflict avoidant or more likely to diffuse problems than other families. Diffusion or avoidance was a recognisable interactional style but was not either common or unique to AN families.

There were procedural differences from Minuchin's study that could account for this disparity. In this study families were given a problem to discuss that had already been elicited from them making it difficult for them to avoid discussing the problem altogether whereas in Minuchin's study the family were in effect asked to "think of a problem".

Le Grange (1989) argued that if the low EE criticism scores he found in his AN families meant that families were well he would have expected higher levels of warmth and positive remarks than he did find. Le Grange did not have a control group. In the present study the levels of warmth and positive remarks in the AN group were indistinguishable from the well families.

10.5.2. Enmeshment and Overprotection

Correlations between "enmeshment" and "overprotection" items on the EFS scale and on the problem solving task were so high that there was no justification for considering them as separate dimensions. AN patients described themselves as being slightly more enmeshed than the other index

children. This was partly but not wholly accounted for by female sex. However, AN families as a whole did not see themselves as more enmeshed than either the CF or the well groups. Nor did they as predicted have lower scores on the Affective Expression and Affective Involvement subscales of the FAM.

Only 2 studies known to the author have attempted to assess directly Minuchin's enmeshment or overprotection concepts in AN families through self report methodology. Using the Leuven Questionnaire, Kog and colleagues (1985) found that AN patients but not their parents showed higher "cohesion" than their well control group which parallels the finding of the present study. By contrast Harding and Lachenmeyer (1986) found no difference in overprotection and enmeshment assessed on the SFIS (see Section 2.10.1) between AN patients and controls. Differences between test items may account for the inconsistent findings.

Two caveats are appropriate. Although the author was at pains to ensure that items included in the EFS satisfactorily tapped Minuchin's descriptions by involving several experienced family therapists in their selection, some of the most "psychosomatic" items did not survive item analysis (see section 9.6.9). Secondly, questionnaires of this kind are indeed self reports only with the implicit limitations as discussed in section 10.1.1 above.

Dare (1991) suggested a conceptual link between the EE measure of emotional overinvolvement and Minuchin's overprotection. In this study this proposal was supported by the emergence after principal components analysis of a factor (factor II) with high loadings on both EE overinvolvement and overprotection as measured on the Problem Solving Task. 74%, 59% and 23% of AN, CF and well families respectively had scores greater than the median, a finding again supportive of Minuchin's observation that enmeshment\overprotection was a feature of AN family style. However although there were similarities between the EE concept and Minuchin's concept there were also important differences.

Minuchin stated (1975) that a necessary condition for the emergence of the "psychosomatic family" was the existence of illness in the child so the finding that EOI was higher in both ill groups in terms of his theory is not surprising. However the EE overinvolvement concept is not equivalent to Minuchin's overprotection concept. The key difference is that the EE concept describes a one way relationship in which the mother and/or father is protective of their child whereas Minuchin described a concept in which the child is just as likely to display nurturing overprotective behaviour towards the adult. The quality of "speaking for" somebody, of knowing what they think or feel could be found in the child as well as the parent. AN families showed more enmeshed/overprotective behaviour than CF and well families, suggesting that the presence of the AN child was the catalyst for enmeshed/overprotective interactions. The scoring system on the problem solving task did not formally identify whether evidence for enmeshment\overprotection came from the child or parent but an examination of the problem solving transcripts shows that there was not a single example of a CF child exhibiting enmeshed behaviour whereas this was common in the AN child.

The second feature of Minuchin's enmeshment concept was the lack of intergenerational boundaries , the process by which a parent or child act in role inappropriate ways. This aspect of enmeshment is absent from the EE concept. Correlations between the generational boundaries measure in the study and the other enmeshment scores were so low that they had to be interpreted as separate dimensions. There were in fact very few examples of weak generational boundaries. CF families were more likely to have well defined boundaries than the 2 other groups. Subjects in this study were older than in Minuchin's study. This may explain why weak boundaries were sparingly rated in this study.

There are no controlled observational studies known to the author that measure "enmeshment" and "overprotection" in AN families to which the present findings can be compared. Kog et al (1985) attempted to measure "boundaries" with a behavioural task (not an observational task) and found

AN families to be less differentiated than well families. (The limitations of Kog's operationalisation of "enmeshment" were discussed in Section 2.10.3.) Rastam and Gillberg (1991) interviewed a parent of AN and well controls and did not find that AN families were more likely to show enmeshment and overprotection. However they did not observe the families in an interaction.

In summary then the findings confirm Minuchin's observation that AN families were likely to be enmeshed and overprotective of each other but not that they were likely to show blurred generational boundaries. However as 56% of AN families did **not** show this pattern it is by no means universal as an interactional style.

10.5.3. Rigidity

> "The rigidity of the anorectic family is not the rigidity of the stone but the ebb and flow of water. The difficulty faced by the therapist is that when he pushes, the family moves."
>
> Minuchin et al, 1978, p.105

The difficulty faced by the researcher is that this is a formidably difficult concept to operationalise. This study failed to overcome the problem of devising a suitable observational task. Minuchin himself in his original work also failed in this endeavour (1978, p 38). In the Belgian study, Kog and colleagues' attempts at operationalisation were inadequate (see Section 2.10.3). In future research observation of a series of therapy sessions might enable the concept to be measured. However, if the concept cannot be operationalised, it cannot be falsified and its utility as a concept must remain in doubt.

In this study the only operationalisation of "rigidity" was the rigidity subscale of the EFS, the items of which were initially chosen by experienced family therapists and subsequently selected by item analysis in the schools project. No differences emerged between households in the 3 groups in this

study. The only difference was that CF patients saw themselves as being more rigid than the 2 other groups. This may reflect a wish for the order and stability that could be helpful in managing a complex disorder such as CF.

10.5.4. The Child's Involvement in Parental Conflict

The fifth characteristic of Minuchin's "psychosomatic family" was that the family had a vested interest in maintaining the child as sick because this served to deflect parental conflict. This hypothesis was not formally tested in this study. However a prediction from this premise would be that improvement in the AN child's health should be followed by an escalation in parental conflict. This prediction was not supported by findings from the self report measures, the EE index or the problem solving task for either the AN or the CF families.

10.5.5. The Psychosomatic Family

Was there support for the idea of the psychosomatic family as a unitary concept? AN families were observed to be both poorer problem solvers and more enmeshed/overprotective than the other groups. However, against prediction there was no correlation between problem solving ability and enmeshment, giving no support to the idea that these 2 dimensions form part of a constellation, the "psychosomatic family".

Also against prediction was the finding that on the self report measures "Enmeshment" correlated negatively with "Conflict" and "Rigidity". In other words, families who saw themselves as enmeshed also saw themselves as flexible and good at resolving conflict. This was also found in the School study.

Were there any psychosomatic families? There were 4 or 5 families in the study who seemed "textbook" cases, 2 of whose behaviour was almost a parody of Minuchin's description. Neither of these 2 families was in the AN

group. Thus it seems recognisable as a family interactional style, albeit a rare one and not typical of Anorexia Nervosa. This was the conclusion too of the Belgian studies (Kog et al 1985, 1989) and the Swedish interview study of AN mothers and well controls (Rastam and Gillberg, 1991).

10.5.6. The Psychosomatic Family and Other Illnesses

Since the publication of Minuchin's book "Psychosomatic Families" (1978), clinicians and researchers have shown interest in the psychosomatic family in illnesses other than Anorexia Nervosa. These have ranged from savage attacks such as Coyne and Anderson's (1988, 1989) hostile critique of Minuchin's original work on the families of poorly controlled diabetics to theoretical attempts to reformulate some of his ideas (eg Wood & Talmon, 1983). There have also been attempts to replicate the findings. There are studies of eczema (Loader et al, 1980), asthma (Burbek, 1979; Gustafsson et al, 1987) and diabetes (Gustafsson et al, 1987). While all these studies have been beset by methodological problems, again the broad conclusion has been that the "psychosomatic family" is a recognisable style but only one of many styles displayed by families.

10.5.7. Changes in Family Functioning

Minuchin and colleagues (1978) claimed that 86% of their AN patients made a full weight and social functioning recovery following the restructuring of their families through family therapy. It would follow from this that improvement in AN family functioning would predict health improvement. In CF families where only a small percentage of families would be expected to be "psychosomatic", the relationship between change in health status and change in family functioning was predicted to be much weaker. It had been predicted that over the 8 month follow-up period there should be a much closer relationship between change in health status and change in family functioning in the AN group than the CF group. However, the relationship between changes in health status and changes in family functioning was not close for either of the ill groups. There were no significant differences in

the strength of that relationship between the AN and CF groups. The methodological limitations of this finding were discussed in 10.1.1.

10.6. The Implications of Dysfunctional Characteristics

It is one thing to observe interactional difference between families. It is another to say what the implications of these characteristics are both for treatment and for prognosis.

In this study negative affective style measured either by self report or observed on the problem solving task or during the Camberwell Family Interview seemed to have no relationship to the health status of CF or AN young people at either baseline or follow-up. This differs from the findings in AN families of le Grange and colleagues (1992b) and van Furth (1991) who both found maternal CC predictive of poor outcome. Paternal CC was predictive in le Grange's study only. No obvious explanation for the difference presents itself in terms of the populations studied or the methodology used, van Furth also having used the Camberwell Family Interview. All le Grange and van Furth's subjects were in active treatment compared to only a proportion in the present study so perhaps criticism may have a effect in mediating treatment efficacy.

The finding of higher level emotional overinvolvement in AN and CF families and its relationship with illness severity is supportive of the supposition that this behaviour style is a natural reaction of parents to grave illness in one of their adolescent children and that it is not a unique feature of AN families.

Differences between the CF and AN families only emerged when the sick child joined the discussion on the problem solving task. In Chapter 8 it was suggested that some of the interactional difficulties of AN families could stem from 2 sources. Firstly, they were dealing with much more difficult behaviour than the CF families. The personality, behavioural and neurotic difficulties of the AN child are in sharp contrast to the psychologically

robust CF young people. Secondly AN families had effectively been deskilled by rejection as collaborators by some treatment agencies. This rejection is widely reported in the popular press usually in personal accounts of patients and their families but for the most part is absent from the academic literature. Chapter 3 reviews the literature on cystic fibrosis. The observant reader may have noticed a section describing research on the burden of illness experienced by CF families. There is no such section in chapter 2 which reviews the AN literature because the problems in AN families are not conceptualised by researchers in this way, the focus remaining on family pathology.

Of course, it can still be argued that it is the behavioural style of the parents that cause the constellation of AN difficulties; this after all is the point of view of Bruch (1978), Minuchin et al (1975) and Crisp and colleagues (1991) among others. Against this view is the finding that many of the established risk factors for psychopathology (reviewed in Chapter 1.5.) are not found more frequently in AN families(reviewed in chapter 2.9.) These risk factors were marital discord, personality disorder, low expectations, social adversity, sexual abuse and physical violence. Although research has established some family transmission both of eating disorders and general psychopathology, because of low incidence most AN patients are not brought up in such households. There is one known risk indicator that does require further examination. In an uncontrolled study, Schmidt and colleagues (1993) found lack of parental care (high indifference, low control) reported by 20% of their AN sample. Their finding is most unusual, for lack of parental care is not reported as a common feature of AN families in the case report literature. More research would be of interest here.

A report by Mischler and Waxler (1968) suggests an interesting methodology whereby some of these ideas could be tested. They observed mothers with a schizophrenic daughter and then separately with a healthy daughter. They found the mothers unresponsive and aloof with the schizophrenic daughter but behaved normally with their well daughter.

Failure to find more dysfunction in AN families than in well families does not of course argue against the possibility that where gross dysfunction exists, prognosis is likely to be poor, and such dysfunction will have to be managed as part and parcel of the treatment package.

What is the way forward on helping AN families? In the context of discussing pitfalls during treatment and detrimental therapist reactions, Vandereycken and Meerman (1984) have a useful commentary on this vexed problem. They remark that many psychiatrists have the attitude that parents and families are the enemy and that when faced with a difficult patient will scapegoat the family. They point out that all families feel some guilt, however unwarranted, but that it could be much more productive to treat families as treatment resources rather than adversaries. This of course was how all the CF families but only some of AN families were treated in this study. AN patients are not generally popular. Fleming and Szmukler (1992) examined attitudes to patients with eating disorders and reported that medical and nursing staff in a general hospital liked these patients less than schizophrenics and saw them as responsible for their illness almost to the same degree as recurrent overdose takers.

In particular, Vandereycken and Meerman warn against the automatic assumption that all AN families need family therapy while acknowledging that this will be useful in some cases. Their comments seem particularly pertinent as 8 out of the 9 families who experienced family therapy in this study found it an unhelpful experience. Researchers are now starting to look at the efficacy of different styles of family work (le Grange et al 1992a,1992b) but would do well to invite commentary from the consumers.

Vandereycken and Meerman also caution against the behavioural treatment approach demanding strict isolation of the patient from the family (no letters, telephone calls, visits), pointing out that the rejection of parents that such a regime implies can of itself elicit a negative reaction in those parents.

The well families in this study did not conform to some gold standard stereotype of normality. A rich variety of interactional styles seemed "good enough". While AN families have the common problem of managing a difficult illness as **families** they too deserve to have their individuality respected and are not well served by unwarranted negative interpretations of their behaviour. Above all these families cannot be ignored. The chronic nature of AN demands that a long term view of family burden is taken and that this planned for with care and compassion.

10.7 Summary and Conclusions

The first objective of the study was to investigate whether any of the characteristics thought to be indicators of family dysfunction were present in the families of AN patients more frequently than in the families of a young person whose illness had a clear cut non-psychogenic origin. While there was no one typical family style and overinvolvement seemed to be related to illness per se the AN families were more likely be poor problem solvers and, in the Minuchian sense, to be enmeshed and overprotective but were no more critical than well families. The second objective was to address the conundrum of whether disturbed families maintain AN over time or whether disturbed interactional styles are no more than a response to changes in illness severity. No significant differences were found between the AN and CF groups although this finding must be interpreted in the context of the methodological difficulties of the follow up study.

While the clinical implications of these findings have been discussed in section 10.6., a number of avenues of further research invite attention. Firstly there could be further developments in research methodology. One of the most satisfactory aspects of this project was finding that with some persistence and focus it was possible to obtain reliable measures of observed family interaction. This study centred on Minuchin's characteristics but there is no reason why other behaviours of clinical interest could not be assessed in a problem solving task or brief family discussion. Secondly the rich variety of family styles in all 3 groups suggest that there may be horses for courses. Treatment trials should where possible include process measures that could help to refine judgements on whether a particular family would benefit from family therapy, for example, or a different approach. Thirdly the success of family therapy with younger, less chronic patients (Russell et al, 1987) has diverted attention away from what is the best way of helping the families of older patients. The efficacy of, say, parent support groups could be compared with family interventions along the lines of the schizophrenia studies (Tarrier, 1989) or compared with individual cognitive behavioural work with the patient on family issues. Fourthly in contradiction to the tradition of random allocation in treatment trials consideration could be given to a study allowing for parental and patient choice into how the family should be managed. Families reported again and again how their wishes were ignored and permitting choice could have some effect on the major AN treatment problem of compliance.

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APPENDIX I: CAMBERWELL FAMILY INTERVIEW (CYSTIC FIBROSIS/ANOREXIA)

Structure of Household

1. Who lives in your household?
   Establish occupation for parents and patient.

   Do you have other children not living at home?

   Have you had any other children who have died?

2. Has anyone else in your family had a problem, with anorexia/cystic fibrosis?

History of Illness

Obtain a brief chronological account of whole history of illness including approximate dates.

3. I should like to begin by asking when X's trouble first began.
   Probes: When did you first notice something different about him/her?
   Was it a sudden or gradual deterioration?
   (Get events) How was behaviour at this time?
   What happened then?
   How long had this been going on?
   How did you feel when this was happening?
   Did it make you feel on edge?

4. What has been the most difficult period for you in managing the illness in this time?

Parental Involvement in Treatment

5. Could I ask you about your role in managing the illness?
   (Much of this will already have been given)
   Probes: Visits alone to GP or Hospital
   Attended family meetings
   Visits at home from NHS
   Visits at home from voluntary agencies
   Attended support groups

Health in the past three months

I should like to ask you in more detail now about how things have been in the last three months.

6. What has X's general state of health been like in this time?

I'd like to ask some questions about the way X may have been affected by this trouble ... and about some sorts of behaviour which people we see sometimes have. Of course, many won't apply, but I should like to run through these quickly and

perhaps you'd tell me whether or not X has been like this, particularly in the last three months.

Omit the disease specific questions as appropriate.

General probes - these probes only need be asked to open up a closed answer.

Onset	When did this first begin?
Severity	How did this show itself?
Frequency	How often?
Reactions	How did you react?
Tension	Did it make you feel on edge?
Coping	How did you deal with it?

Try and assess degree of parental involvement.

7. What has X's sleep been like?
   Probe: Has he/she had any difficulty in getting off?
   Nightmares? Waking up early?

8. Has X complained of any aches or pains?

9. Has X been lacking in energy? Or seemed particularly tired?
   Probe: Sitting around not doing much?
   Has he/she stopped doing anything?

10. Has he/she seemed overactive at all?
    Probe: Restless? Exercising? Agitated?

11. Has he/she had any problems with concentration?

12. Has he/she hurt anyone or hit anyone?
    Probe: Can you describe what happened?

13. Has he/she had any unusual fears? Periods of anxiety?

14. Has he/she been depressed at all?
    Probe: What makes you think he/she was miserable?
    Has he/she tried to harm herself at all?

15. Could you describe/what problems have there been with X's eating?
    Probe: Dieting? Fussy? Special diets? Overeating?
    Do you prepare special foods for him/her?

16. Has there been any problem with vomiting?

17. Have there been any problems with X's bowel functioning?
    Probe: Laxatives? Foul stools? Constipation?

18. Have/What problems have there been with X's chest?
    Probe: Cough? Physiotherapy? Infections? Effort?

19. Have there been any problems with medication?
    Probe: Does he/she take pills regularly?

Are you involved in supervising the taking of tablets?

20. Have there been any other worrying or trying symptoms or behaviour that we haven't touched on?

Parental Involvement with Agencies

I want now to ask you in the past three months what contact have you yourselves had with the hospital or support services.

21. Do you accompany X to medical appointments?

22. Have you been to speak to anyone at the hospital or to your GP on your own about the illness?

23. Have you attended any family meetings?

24. Home visits from NHS people?

25. Home visits from voluntary agencies?

26. Been to any voluntary groups?

Family Time Budget

27. I'd like to get a picture of how X usually spends a day. On a usual weekday how would the day start?
(Get a picture of the average day)

28. And at weekends?

Irritability

One of the ways in which being unwell can affect people is to make them more irritable, I mean snappy, or more likely to fly off the handle at little things.

29. How often has X been like this with yourself?

If denied ask: Has he/she been cross at all?
Can you remember a time when he/she lost his/her temper in the past year?

30. Can you describe what went on the last time this happened?

31. Can you cope with the irritability at all?

Relationship with Child

Can you tell me a bit more about how you and X get along?

32. Do you find him/her a friendly person?

33. Is he/she easy to get on with?

34. Can you get close to him/her?

35. In what ways would you like him/her to be different?

36. In what ways does he/she get on your nerves?

37. Is X interested in boys/girls at all?
Probe: What do you feel about the future in that respect?

Family Strain

38. What difference has your child's problem made to you and the family?

39. From your point of view, what has been the most disturbing aspect of your child's problem?

40. In the past three months have there been/what has been the main strain?

Satisfaction with Management

41. Are you satisfied with the way X is being cared for/managed by the health service?

42. What changes would you like to be made? What extra help would you like?

END OF INTERVIEW. ASK CHILD TO COME IN.

PARENTS AND CHILD INTERVIEW

This section of the interview was not rated for Expressed Emotion but was used as a bridge before the family began the problem solving discussion.

Family Relationships

43. How would you describe yourself as a family?
What words would you use to describe yourselves as a family?
If the family replies, follow this up. If not then:

44. Families vary a lot in the way their members get together. Some are together most of the time, others tend to do things separately. How does your family fit on this wide range?

45. Do you do things together as a whole family?
Probe: Eat together: Watch TV together?
Who does what with whom?

46. How do you tend to organise meals at home?

47. What about contact with people outside the home. Do you see much of other relatives?

48. What about social life outside the home?
Probe: Friends, local groups, clubs?

49. What about X? Does he/she spend much time outside the home?

Quarrels

50. Most families have quarrels or arguments from time to time. Apart from the sort of irritability we have been talking about, what sort of things does your family have disagreements about?

If a spontaneous response is made

51. How do the disagreements happen? Can you tell me what it is like?

52. Has X had any fights or quarrels with brothers and sisters?
Probe: Do they call each other names?
Do they shout at each other?

53. Parents and children often argue about things - what about you?
Do you ever have periods of not speaking after a disagreement?
How do disagreements happen?
What is it like?

Use appropriate probes:
Bedtime?
TV time?
Drinking?
Smoking?
Friends?
Staying out late?
Clothes?

54. Could you describe to me the last time you had a quarrel with X?

55. Husbands and wives also have their differences - do they occur in front of the children?
Probe: Managing the children?
Money matters?

Household Tasks

56. Various jobs have to be done in the family? Who does what in your household?
Probe: Who tends to do the shopping?
And the cooking?
Repairs to the house?
Money matters/Bills?
Who sees to the children?
Are their any specific jobs for X?

57. Is this a fixed pattern? Have you ever considered whether you wanted to change the way things are done?

58. Are you satisfied with these arrangements?
Probe: Do any of these things lead to disagreements?

APPENDIX II: CAMBERWELL FAMILY INTERVIEW (WELL)

Structure of Household

1. Who lives in your household?

2. Do you have other children not living at home?

3. Have you had any other children who have died?

4. Are you working? What about X?

History of Illness

5. Has anyone in your family had an eating problem?

6. Have you had any major health problems?
   Ask of both parents.

7. Have you had any trouble with your nerves - any psychological troubles?

8. What has X's health been like over the years?
   If any illnesses are mentioned, probe for events and attitudes.

9. Many families have a spell when they find bringing up a particular child trying. Looking back over the years, what has been the most troublesome period for you?

10. How have you found the teenage years?

11. How have things gone at school?

Health in the last three months

I'd like to ask some questions about X's health in the last three months. Of course, many won't apply, but I should like to run through these quickly and perhaps you'd tell me whether or not X has been like this, particularly in the last three months.

Omit the disease specific questions as appropriate.
General probes - these probes need only be asked to open up a closed answer.

Onset	When did this first begin?
Severity	How did this show itself?
Frequency	How often?
Reactions	How did you react?
Tension	Did it make you feel on edge?
Coping	How did you deal with it?

Try and assess degree of parental involvement.

12. What has X's sleep been like?

Probe: Has he/she had any difficulty in getting off? Nightmares? Waking up early?

13. Has X complained of any aches or pains?

14. Has X been lacking in energy? or seemed particularly tired?
Probe: Sitting around not doing much? Has she stopped doing anything?

15. Has she seemed overactive at all?
Probe: Restless? Exercising? Agitated?

16. Has she had any problems with concentration?

17. Has she hurt anyone or hit anyone?
Probe: Can you describe what happened?

18. Has she had any unusual fears? Period of anxiety?

19. Has she been depressed at all?
Probe: What makes you think she was miserable? Has she tried to harm herself at all?

20. Could you describe/have there been any problems with X's eating?
Probe: Dieting? Fussy? Special diets? Overeating? Do you prepare special foods for her?

21. Has there been any problem with vomiting?

22. Have there been any problems with X's bowel functioning?
Probe: Laxatives? Foul stools? Constipation?

23. Have/what problems have there been with X's chest?
Probe: Cough? Physiotherapy? Infection? Effort?

24. Have there been any problems with medication?
Probe: Does he take pills regularly? Are you involved in supervising the taking of tablets?

25. Have there been any other worrying or trying symptoms or behaviour that we haven't touched on?

Family Time Budget

26. I'd like to get a picture of how X usually spends a day. On a usual weekday how would the day start?
Get a picture of the average day.

27. And at weekends?

Irritability

Everyone gets short tempered or irritable from time to time, or more likely to fly off the handle at little things.

28. How often has X been like this with yourself?
If denied ask: Has he been cross at all? Can you remember a time when he lost his temper in the past year?

29. Can you describe what went on the last time this happened?
Probe for X and all members of the household.

30. Can you cope with the irritability at all?

Relationship with Child

31. What sort of a person is X and how do you get along?
Probe: Is he/she a friendly person? Can you get close to her? Is she easy to get on with?

32. In what ways does she get on your nerves?

33. In what ways would you like her to be different?

34. Is X interested in boys/girls at all?

END OF INTERVIEW. ASK CHILD TO COME IN.

PARENT AND CHILD INTERVIEW

This section of the interview was not rated for Expressed Emotion but was used as a bridge before the family started the problem solving discussion.

Family Relationships

35. How would you describe yourselves as a family? What words would you use to describe yourselves as a family? If the family replies, follow this up. If not then:

36. Families vary a lot in the way their members get together. Some are together most of the time, others tend to do things separately. How does you family fit on this wide range?

37. What about X? Does he tend to do things with you?

38. Do you do things together as a whole family?
Probe: Eat together? Watch TV together? Who does what with whom?

39. What about contact with people outside the home? Do you see much of other relatives?

40. What about social life outside the home?
Probe: Friends, local groups, clubs?

41. What about X? Does he/she spend much time outside the home?

Quarrels

42. Most families have quarrels or arguments from time to time. Apart from the sort of irritability we have been talking about, what sort of things does your family have disagreements about?

If a spontaneous reponse is made:

43. How do the disagreements happen? Can you tell me what it is like?

44. Has X had any fights or quarrels with his brothers and sisters?
Probe: Do they call each other names? Do they shout at each other?

45. Parents and children often argue about things? What about you? Do you ever have periods of not speaking after a disagreement? How do disagreements happen? What is it like?
Use age appropriate probes: Bedtime?
TV time?
Drinking?
Smoking?
Friends?
Staying out late?
Clothes?

46. Could you describe to me the last time you had a quarrel with X?

47. Husbands and wives also have their differences. Do they occur in front of the children?
Probe: Managing the children. Money matters.

Household Tasks

48. Various jobs have to be done in the family? Who does what in your household?
Probe: Who tends to do the shopping? And the cooking? Repairs to the house? Money matters, bills? Who sees to the children? Are there any specific jobs for X?

49. Is this a fixed pattern? Have you ever considered whether you wanted to change the way things are done?

50. Are you satisfied with these arrangements?
Probe: Do any of these things lead to disagreements?

APPENDIX III: THE PROBLEM SOLVING TASK

Objectives

The objective of the problem solving task was for the family triad of mother, father and index child to discuss an issue or problem salient to their household and for this interaction to be assessed for presence of some of the concepts that Minuchin measured in his original work.

The Family Task

As a medium for measuring the concepts of enmeshment, overprotectiveness, low tolerance of conflict and rigidity, Minuchin asked his families to carry out a family task (Minuchin et al, 1978). This was an elaboration of the Wiltwyck Family Task (Minuchin et al, 1967) and is described in Chapter 2, Section 2.10.3.

Reviews of family interaction tasks (eg Markham and Notarius, 1987; Jacob and Tennenbaum, 1988) invariably conclude with the advice that tasks that reflect problem-solving situations such as a discussion of that family's key problem are preferable to less salient tasks such as sorting cards because a family's ability to solve real-life problems is critical to adaptive functioning. The first challenge was to devise a satisfactory family task.

Development

In order to focus more sharply on a meaningful family task and to deal with the limitations of the Philadelphia Child Guidance Clinic scoring guide, pilot work was carried out on 5 families not included in this study. Minuchin's family task (1978), Kinston and Loader's family task interview (1988), Kog and Vandereyken's conflict task (1985, 1989) and the revealed differences technique using the Inventory of Parent Adolescent Conflict (Olson et al, 1978) were methodologies tried out but rejected for the following reasons: low level of acceptability from the families, too long, more appropriate for a younger age group or a task so remote from a family's own difficulties that families remained disengaged from any affective behaviour. The most satisfactory task, the one adopted for this study, was based on procedures used by Doane (1981) with the families of young schizophrenic patients.

The procedure was as follows. At Interview 1 with the index child, the independent assessor used a semi-structured interview, the cue elicitation procedure (see below for copy) (Doane, 1981) to elicit areas of family conflict. At Interview 2 family conflicts described by parents were identified during the Camberwell Family Interview. Doane also used a cue elicitation procedure for parents but that was not used here. From these 2 sources 2 problems idiosyncratically relevant to that family were selected for discussion in the Problem Solving Task. Issues directly related to the index illness were if possible avoided. At the end of Interview 2 the family triad were given the following instructions:

> "During the interviews several areas of differences with the family have come to light. There is nothing unusual about this - no family agrees on everything. I am going to describe to you a situation that you have told me about. I would like you to discuss it together, say what you feel about it and see if you have any suggestions you want to make about it. I will give you ten minutes to talk together. While you are doing this, I shall leave the room although the tape recorder will be running. If ten minutes is too long, then come and get me."

At this point the interviewer described the first problem, turned the tape recorder on and left the room. She returned after 10 minutes and the procedure was repeated for Problem 2.

Examples of problems presented to families for discussion were as follows:

> "Veronica, you told me earlier that you felt that your brother got treated more favourably by your father than you did and that this wasn't fair. Mrs Jones, you had told me that Veronica always seemed jealous of her brother."

> "Mrs Smith, when I asked how you would like Jimmy to change, you said that his room was a tip and you wished he would look after it. Jimmy, you told me that you were fed up with your parents going on and on at you about your room."

The Scoring System

Background

The second challenge was to devise a rating system that was both reliable and valid in that it would satisfy the demands of both the clinican and the statistician. Describing the history of family interaction research, Eisler and colleagues noted that the greater the researchers' efforts to resolve the methodological problems of developing precise operationally defined measures of family interaction, the less relevant their efforts seemed to be to clinicians (Eisler et al, 1988).

There have been successes and failures in this endeavour. Loader et al (1980) attempted to rate the "psychosomatic family" in families containing a child with atopic eczema using the Current Family State Assessment (Kinston et al, 1979). This consists of 33 items of interaction, each of which is scored on a 5-point scale, some of which are directly relevant to Minuchin's hypothesis. They achieved only modest agreement between raters but it has to be pointed out that the raters were set a formidable task. They were asked to make global ratings after watching an hour long family interview. The CFSA itself has only minimal guidance on anchor points and lacks examples of the kind of behaviour that would warrant high or low ratings.

Eisler et al (1988) pointed out that the Expressed Emotion Index is an example of an instrument that has successfully combined a considerable degree of judgment about specific behaviour *(thus giving clinical flexibility)* with precise operational definitions *(that would permit good inter-rater reliability).*

Eisler et al (1985) showed that observers could be trained to recognise complex and idiosyncratic descriptions made by other clinicians about a particular family with a high level of discrimination. Street and Foot (1984) found that they could train family therapists to recognise structural concepts from Minuchin's theory by matching brief video-clips of family interaction to written vignettes of each structural concept.

In the past there have been several attempts to devise a scoring system to quantify problem solving skills (see Jacob and Tennebaum, 1988 for review). One of the

most satisfactory of these is the Kategortensystem fuer Partnerschaftliche Interaktion (KPI) (Hahlweg et al, 1984) which has the merit of describing categories of communication skills that seem both clinically meaningful and are operationalised in a scoring manual of Teutonic thoroughness. A disadvantage of the KPI is that it uses a micro-analytic approach to scoring which is both tedious and labour intensive (24 hours per case). This disqualified the scale from use in the present study. However, examples of problem solving strategies found in the KPI manual were incorporated into the Problem Solving Task manual to assist in the assessment of problem solving skills.

Thus it seems that if the rating task is brief and well defined, the scoring manual explicit, and opportunities for recognition maximized, it should be possible to achieve agreement between raters. Drawing on these ideas and wherever possible from the Philadelphia Child Guidance Clinic manual a scoring manual was produced.

Transcripts were made of the audiotapes and ratings were made of several "psychosomatic" dimensions. The dimensions were: alliance shifts, mind reading, weak generation boundaries, mediating responses and closeness (all enmeshment concepts), overprotection and resolution of conflict. This study failed to operationalise rigidity - as had been the experience of Minuchin.

Ratings on the problem solving task were carried out by the author. As she was blind to neither the objective of the study nor the patient groups, a sample of 20 tapes stratified for subject group was rated blind by an independent assessor[1]. Kappa co-efficients were good to very good on all dimensions apart from "closeness" - see Table III.1.

[1] Ms Maisie Gard, Research Psychologist, University of Edinburgh

TABLE III.1 PROBLEM SOLVING TASK - AGREEMENT BETWEEN RATERS

n = 20			
Dimension	kappa statistic	95% confidence interval	Strength of agreement
Problem solving task	.84	.63 to 1.0	Very good
Alliance shifts	.85	.57 to 1.0	Very good
Gen. Boundaries[1]	.68	.28 to 1.0	Good
Mind reading	.66	.34 to .98	Good
Mediating response[1]	.93	.81 to 1.0	Very good
Closeness	.46	0 to 1.0	Moderate
Overprotection	1.0	-	Perfect

[1] weighted kappa

Correlations between the dimensions are shown in Table III.2. The high correlation between overprotection and most of the "enmeshment" concepts did not justify describing it as a separate concept. By contrast generational boundaries did not correlate with the enmeshment concepts and was best seen as a separate dimension.

TABLE III.2 PROBLEM SOLVING TASK DIMENSIONS CORRELATION BETWEEN SCALES n = 83

	ALLI	MIND	MED	GEN	CLOSE	OVER
ALLI	1					
MIND	**.27**	1				
MED	**.36**	**.29**	1			
GEN	.17	-.03	-.12	1		
CLOSE	-.03	**.27**	-.17	-.04	1	
OVER	-.03	**.33**	-.08	-.12	**.30**	1
CONF	.05	-.07	-.01	-.13	.11	.06

Spearman's r: missing data pairwise deleted
All correlations where $p < .03$ in bold

KEY

ALLI	Alliance shifts
MIND	Mind reading
MED	Mediating response
GEN	Weak generation boundaries
CLOSE	Closeness
OVER	Overprotectiveness
CONF	Poor resolution of conflict

Were the objectives of the Problem Solving Task met?

Efforts to ensure that the problems under discussion were salient to that household paid off in that most families sounded as if they were engaged in a real discussion. In only 4 out of the 85 families who carried out the problem solving task did the discussion sound artificial.

A number of ingredients contributed to the success in achieving good inter-rater reliability for most concepts. The task was short (10 minutes per problem) and both transcript and audio-tape were available to the rater permitting vigilance to be maximised. Both written descriptions of Minuchin's concepts and examples of the kind of behaviour that constituted evidence were available to the independent rater. She also had worked examples of tapes and ratings to train on and close supervision in the training period. Thus there was plenty of opportunity for her to recognise examples of "psychosomatic" behaviour.

CUE ELICITATION PROCEDURE (Doane, 1981)[2]

Cue Elicitation Interview

1. Most people have things that they like best about their family as well as things they don't like. What sorts of things do **you** think go rather **well** in your family? (eg experiences, activities and so on)

2. Okay, fine, now how about some of the things that don't go as well as you'd like, or things you'd like to see changed?

AT THIS POINT INTO THE INTERVIEW IT IS UNLIKELY THAT YOU WILL BE ABLE TO OBTAIN TWO GOOD CUE STATEMENTS FROM THE PATIENT. HOWEVER, WITH SOME PATIENTS YOU MAY BE ABLE TO ELICIT YOUR CUES FROM QUESTION 2. IF NOT, GO ON THROUGH THE FOLLOWING QUESTIONS. YOU MAY SKIP QUESTIONS AT YOUR DISCRETION.

3. You know every family has its rules but not everybody always agrees with **all** the rules. How is it in your family?

   PROBE:

   What kinds of rules do people have in your family?
   Who makes up the rules? For instance, do your parents want you to pick up your clothes or help with chores and things like that?
   Do they want you to be in by a certain time?
   Do they want you to keep the music down?
   Do you ever think there are too many rules?
   Are the rules in your house the same for everybody? eg are they the same for you and your brothers and sisters.

4. In all families people occasionally do things that get on other people's nerves.

   What do your parents do that tend to bug you the most? (Do you do things that get on their nerves?)

   PROBE:

   Do you ever feel that your mother or your father nags you or yells at you about anything, bugs you about anything? What's it like?
   Do they ever make demands on you that you feel you can't quite handle? Or do they ask you to do things that you don't feel you should have to do, like get a job, go to school, do chores or getting out and meeting people and so on, or getting up too early?

   Do you ever feel that they may be unreasonable, like asking you to take care of things around the house that you don't feel are totally realistic or

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[2] Obtainable from:
Dr J.A. Doane, Yale Psychiatric Institute, P.O. Box 12A, Yale Station, New Haven 0652, U.S.A.

necessary - help to prepare meals, cleaning, yardwork, taking out the trash and so on?

5. Do you think your parents want you to spend more time at home than you want to? Or do you think they'd like you to spend less time at home?

   PROBE:

   Do you think they spend enough time with **you**?
   If no, would you like your mom and dad to spend more time with you?

6. Do you ever get the feeling that your parents worry too much about you, about going out and being with your friends and so on?

   PROBE:

   Do they worry about what you do with your friends?

7. Do you and your mom and dad ever get into hassles about money?
   What usually happens?

8. Can you think of anything that you do that maybe bothers your parents a little bit or bugs them or gets on their nerves?

   PROBE:

   You know, like drinking or smoking or anything like that?

9. Do your parents ever ask you to do things that you think are really none of their business? (eg like taking a shower, brushing your hair, changing your clothes, or something like that).

   PROBE:

   Do you ever feel that they may be asking you too much or too often, or do you wish they would not do it?

10. Do you ever feel that your parents want to talk to you about something and you really don't feel like talking about it; you'd like to keep it to yourself?

11. Do you ever feel the opposite way, maybe you have some thoughts that you'd like to keep to yourself and your parents want you to tell them about it?

12. Would you say it is easier to talk with your mom or your dad?

    PROBE:

    What kinds of things make it harder to talk to your mom/dad? (ie whichever one is hardest to communicate with)

13. Do you prefer being out with people most of the time, or do you usually prefer more to be alone, perhaps at home?

If prefers to be alone or at home more

PROBE:

Do you feel that your mom and dad push you to get out and be with people before you're ready? In other words, do your parents try to get you to go out more than you feel up to or do they bug you to go out and meet people?

14. You know, one of the things about living in a family is that you, **your friends** and your parents often have feelings _____ about them - sometimes they like them and sometimes they don't. How is it with you?

PROBE:

Do your parents like your friends?
Do they ever criticise your friends at all?

15. Sometimes it's hard to meet new friends. Do your parents sometimes wish you had more friends than you feel you can make?

AVOID THESE TOPICS

Doctor/therapist
Medication
Hospital/ward/staff issues
Eating issues

SCORING GUIDE - PROBLEM SOLVING TASK

While listening to the audiotape, mark on the transcript any examples of mind reading, mediating response, closeness, alliance shift, overprotectiveness, weak or strong generation boundaries. Also all behaviour as described in the manual that will assist in making the categorisation on problem solving skills.

Scores are made from transcripts and audiotapes of the two 10 minute problem solving family discussions.

ENMESHMENT/OVERPROTECTION

Enmeshment refers to a tight web of family relationships in which family members are highly sensitive to each other, wrapped up in each other and very dependent on each other.

In an enmeshed family there is little opportunity to be an individual. There is a lack of privacy. Family members tend to intrude on each other's thoughts, feelings and activities; to "know" what the others are thinking or feeling and to speak for them (mind reading). A family member may relay messages from another family member to a third, blocking direct communication. There may be many interruptions with one family member finishing another's sentence (mediating response).

The needs of the individual take second place to perceived overall family needs. There can be an emphasis on togetherness and "sharing". The family often share common beliefs and opinions and show little overt disagreement. At interview "togetherness" may literally be observed. For example, they may sit very close to each other, share private jokes or whisper into someone's ear making the interviewer feel excluded (closeness).

However, if there is a conflict, then there are likely to be sudden "alliance shifts". A dyadic conflict may set off a chain of shifting alliances with the whole family as other members get involved. An alliance happens when A joins B against C in a discussion. An alliance shift happens when, for example, B then would suddenly shift to defend C against A although the topic under discussion has not changed.

Overprotectiveness

In overprotective families family members show a high degree of concern for each other's welfare. This concern is not limited to the identified patient or to the area of illness. Nurturing and protective responses are constantly elicited and applied as family members interact. A sneeze sets off a flurry of handkerchief offers: complaints and queries about fatigue or discomfort punctuate the flow of communications. Family members' perceptions of each other are structured around probable concerns, particularly where there is a sick child. In such families overprotectiveness can retard the children's development of autonomy and competence. In turn the children, particularly the ill child, feel great responsibility for protecting the family. For the sick child the experience of being able to protect the family by using his symptoms may be a major reinforcement for the illness. Critical remarks are often accompanied by pacifying behaviour. Only two examples of overprotectiveness are given in Minuchin's book. One in which the son asks for a kleenex, the father offers his handkerchief, mothers gives a kleenex to father and daughter offers her handkerchief. The second in which a

child describes an event in the past when she felt her sister had been unjustly punished, ran upstairs crying and asking for the sister to stop being punished.

Scoring

The enmeshment/overprotection score is the sum of mind reading, mediating response, closeness, alliance shifts and overprotectiveness. Each of these dimensions is rated on a 3 point scale: clear evidence, some evidence, no evidence.

1. Mind Reading

Family member says what another person is thinking, feeling or wanting without that person having expressed an opinion. The implication is there of knowing what the other person wants.

Examples:

(a) Daughter has said nothing. Father: "You think your mother keeps herself to herself too much, don't you?"
(b) "Potatoes. You like potatoes."
(c) "I know why she wants to leave hospital. I have tickets for the ballet and she wants to go to the ballet."
(d) "You must feel you've had far more to worry about than anyone else."
(e) "You drink because you enjoy it. You don't drink because it gives you a kick or you feel high or to make you feel on top of the world."

Clear evidence of mind reading	2
Possible evidence of mind reading	1
No evidence	0

2. Mediating Response

These are responses made by a person acting as a pathway between two other people. They replace direct communication between two people. Some mediating responses also involve mind reading. Do not score the same event twice.

Examples:

(a) Mother to father: "He likes potatoes". Father: "Will he eat them?" Mother: "Yes, he will."
(b) Jane: "I am listening." Father: "Jane has switched off". Mother: "Yes, Jane has switched off because she doesn't like it when she realises she can be at fault".
(c) Son (talking to mother, father present): "He (father) always leaves me to do this and doesn't consider some of the things I would like to do".
(d) Mother speaking to father (son present): "You don't believe in the fact that if Jimmy wants to sit and watch television for an hour during the day, you don't think he should do that and I think that's when he gets annoyed". Father: "Yes, if he's going to want to watch things, then fine but he's got to get out and do other things".
(e) Mother speaking to father (daughter present): "I think that now she's got to her age that you are directing her too much".

Clear evidence of mediating response	2
Possible evidence	1
No evidence	0

3. Closeness

If any of the following "togetherness" behaviour is observed, then rate for presence of closeness.

(a) Handling: touching, patting, brushing hair etc.
(b) Closing the gap: looking closely into someone's face, moving one's seat closer, whispering into someone's ear etc.
(c) Joint affective reactions: the family have identical affective reaction, eg crying in unison, laughing together at private jokes etc.
(d) "Sharing" statement: often a "we" statement is used where an "I" statement would be more appropriate.

Examples:

(a) Father is talking about applying for a new job. "You knew before Mum that we didn't get what we wanted last time. We're not really sure what will happen in the end. We'll all be together, sometime."
(b) Father: "We just feel very hopeful for the future".

Clear evidence of closeness	2
Possible evidence	1
No evidence	0

4. Alliance Shifts

An alliance occurs when A joins B against C.

Example: Father is aggressive towards daughter. Mother joins in and supports father against daughter.
or: Father makes a suggestion. Son rejects it. Mother supports son's rejection.

An alliance shift occurs when there is a sudden swing from one position to another.
Example: Mother and father are both critical of daughter. Mother suddenly shifts to defend daughter against father.

One or more shifts of alliance	2
Possible shift	1
No evidence of shift	0

5. Overprotectiveness

If any of the following behaviours are observed, then rate for the presence of overprotectiveness.

(a) Person appears to solicit protectiveness by crying, complaining of hunger, fatigue, headache.
(b) The discussion shifts from the problem the family are meant to be discussing to concerns about feelings, worries, illness etc.
(c) "Comforting" behaviour.
(d) Concern with or easily affected by signs of stress or distress, physical well-being of others.
(e) Statement of overprotective attitude.

Examples:

(a) "I don't want you to be worrying all the time about me"
(b) Mother of 21 year old girl: "I've never thought that I should share my innermost thoughts with my children because I don't really think you should overload them with anything." Daughter: "That's the way I think about you."
(c) Father: "Sally, without listening to what you are saying, we can be aware just from the tone of your voice and just the sound of a conversation that you are upset about something and when you are upset we tend to be concerned about why you are upset ..."
(d) Father: "Do we aggravate you?" Daughter: "No you don't". Mother: "Do you think everyone is bad to you?" Daughter: "I worry about you too in your situation at the moment".
(e) Mother: "Darling, I've upset you. I'm sorry. I'm sorry, I'm sorry".

Clear evidence of overprotectiveness	2
Possible evidence of overprotectiveness	1
No evidence	0

WEAK GENERATIONAL BOUNDARIES

When the differentiation between parent roles and child roles is not always clear, this is described as weak generational boundaries. Children can behave like parents and vice versa. Executive hierarchies are comprised.

For example, children may join one parent in criticising the other. Children may take inappropriate parental roles towards each other. A parent may enlist the support of a child in struggles with the other parent or work at cross purposes in relation to the children. The child may take responsibility for parenting. Family members tend to be the same emotional age.

Examples:

(a) Daughter: "I compare my mother to other wives, and when I compare her, she doesn't fit in. Other wives stock up ... I try to get her to save money but in my opinion she doesn't shop wisely."

(b) Mother (talking about the discipline of her other daughter): "Our measures are strong but Jane is a very strong willed girl". Daughter: "You just have to be stronger-willed than her". Mother: "Yes we do". Daughter: "You can't be ruled by her". Mother: "No, we're not ruled. Do you feel you're ruled by her?" Daughter: "Not at all. I have more control over her than you do."

Good example of parent acting like child or vice versa	2
Possible evidence	1
No evidence	0

RESOLUTION OF CONFLICT

Background

A global judgment is made of each problem discussion which is then allocated to one of three response style categories: A, B or C.

The three categories are based closely on the categories outlined in the Philadelphia Child Guidance Clinic scoring manual (Minuchin, 1978). Categories A and B represent unsuccessful problem solving strategies and category C successful strategies. There have been several attempts in the past to delineate successful and unsuccessful problem solving strategies. To augment Minuchin's descriptors, some of the KPI (Hahlweg et al, 1984) key behaviours to watch for are included as they have proved useful aids in arriving at the global judgment for each category.

Category A - No conflict emerges or conflict diffused

Each family has been asked to discuss a problem which has emerged in interview as being salient to that household. Therefore, it is unusual but not unknown for "no conflict to emerge". The occasional family denies the existence of a problem it has previously acknowledged. A more likely scenario is the family who attempt to diffuse the conflict by brushing it all under the carpet. Tolerance for conflict is very low. One family member may bring up an area of difficulty but another manages to detour to avoid confrontation. Confrontation involving differences of opinion and issues of autonomy and control are avoided or diffused. Consequently there is often a chronic state of submerged, unresolved conflict.

In allocating an "A" rating, look out for the following:

1. Evidence of DENIAL

   (a) The family denies the existence of any conflict.
   eg: "I can't think of anything."
   "There's nothing wrong."

   or

   (b) As soon as one opinion emerges there is instant agreement and consensus.
   eg: "I wish you would clear your room more often." "You're right, I should. I'll try to do it more often."

2. Little evidence of "SELF DISCLOSURE" (KPI)

   (a) Direct expression of feelings.
   eg "I'm too angry to listen at the moment".

   (b) Direct expression of wishes and needs.
   eg "I'd like to go fishing tomorrow".

   (c) Direct expression of attitudes, opinions and behaviour.

3. Little evidence of "PROBLEM DESCRIPTION" (KPI)

   (a) Neutral descriptions of the problem.
   eg "I think we've got a problem with the kids".

   (b) Neutral questions.
   eg "Did the car break down yesterday?"

   (c) With neutral tone.
   eg "I don't think that women should work outside the home".

   The result of this is little clarification of the problem.

4. Evidence of DIFFUSION

There are hints of conflict but they are quickly diffused by any of the following methods.

(a) Suppression

After appropriate or minor disagreement is expressed or conflict is described, pressure is applied. There might be an attempt to retract critical comment.
eg "You've got the wrong impression".

eg Husband expresses criticism of daughter. Wife responds "Remember we all have to try and agree" or "Don't let's have a fight now".

(b) Minimising

Reducing the importance of any issue. Minimising difference between two positions or saying things are the same when they are different.
eg "That's not important, that's just a little thing."

(c) Undoing

A critical remark is diffused by the speaker retracting it or saying nurturant things about the recipient.
eg "I wish you wouldn't come into my room without knocking".
"I don't mean that really.
"It's lovely to see you at any time."

(d) Giving up

At the least sign of opposition or difference of opinion to avoid further conflict.
eg "Yes, you're right. Let's leave that now."

(e) Change of subject

An irrelevant subject is discussed to avoid the task in hand.

<u>Category B - Open conflict - unresolved</u>

This allocation is made to those families who openly disagree with each other but whose poor problem solving skills ensure that no development or movement towards resolution takes place during the problem discussion. There will be a lot of repetition, bickering or oppositional behaviour. Few suggestions will be made as to how to resolve the problem or suggestions made will be immediately rejected out of hand or self justified by one or more of the other parties without any "give". Look out for:

1. High levels of SELF DISCLOSURE (KPI) and of PROBLEM DESCRIPTION (KPI).

2. A high level of "JUSTIFICATION" (KPI):

   (a) Either excusing one's own behaviour, eg "I had a lot of things to do yesterday" or
   (b) Denial of responsibility, eg "That's not my job".

3. A high level of DISAGREEMENT (KPI):

   (a) Direct disagreement, eg "No, that's not true".
   eg "I don't believe what you just said".
   (b) Yes-but, eg "Yes, you're right, but we don't have the money".
   (c) Short disagreeing statements, eg "No".
   (d) Blocking and interruptions, eg "Stop it, I've had enough".

4. A high level of CRITICISM (KPI):

   (a) When the speaker's intention is to hurt, demean or embarrass in a global way, eg "You're lazy".
   (b) When the speaker expresses his or her dislike of a specific behaviour of the listener, eg "The car broke down because you forgot to take it to the garage".

5. A high level of NEGATIVE SOLUTION. The speaker describes something he would like the other not to do in order to solve a problem.
   eg "You shouldn't bring work home".
   eg "You demand too much of your son". "Well then I won't talk to him any more."

Category C - Resolution

The problems that families are asked to describe are often problems of long standing and which the families have in some sense adapted to. Therefore, in the space of a ten minute discussion, it is unlikely that they will have completely worked out a satisfactory solution of their own so, in order to gain a category C score, one of three conditions must be met.

1. Some movement in the discussion towards resolution or some suggestion of compromise.

2. Those situations where a family discussion of a problem leads to a clarification of what the main issues are and then to a realisation that what had seemed like a major problem is in fact not really a problem. (This is different from a family who would be given a score of 2 (minimisation) because that is given where there is no clarification of the problem.)

3. Situations where people agree comfortably to "agree to differ". Most "disagreement" conclusions will get a Catetory B Score so this will only happen where there is a high level of AGREEMENT, ACCEPTANCE AND PROBLEM CLARIFICATION (KPI).

Look out for:

1. A high level of SELF DISCLOSURE (KPI) and PROBLEM DESCRIPTION (KPI).

2. A high level of POSITIVE SOLUTION (KPI)
either

   (a) specific constructive proposals
   eg "I'll do the dishes" or

   (b) compromise
   eg "If you do x, I'll do y".

3. A high level of ACCEPTANCE OF THE OTHER (KPI). Acceptance is of three kinds:

   (a) Paraphrase
   eg "You're saying that the kids are too young to go to kindergarten".

   (b) Open questions are characterised by showing interest and asking well targetted questions about the feelings and internal state of the partner.

   (c) Positive feedback
   eg "I like the way you started this discussion".
   eg "Do you think I react too harshly in such situations?"
   eg "Are you still sad about it?"

4. There is likely to be a high level of AGREEMENT (KPI)

   (a) direct agreement
   eg "Yes, that's right".

   (b) acceptance of responsibility
   eg "I know I started the fight". (Watch out here for conflict diffusion).

SCORING Resolution of Conflict

Each problem discussion is allocated to one of three categories:

(A) No conflict\conflict diffused
(B) Open conflict
(C) Resolution of conflict

This is done for both problem 1 and problem 2.

Resolution of conflict can then be scored in three ways:

1. Good - Poor problem solving skills.

   Score 1 = Good problem solving skills = C + C
   Score 2 = Intermediate problem solving skills = A + C or B + C
   Score 3 = Poor problem solving skills = A + A or B + B or A + B

2. Low - High use of diffusion.

   Each family is then given a "diffusion of conflict score":

   1 = No use of diffusion = C + C or B + C or B + B
   2 = Some use of diffusion = A + B or A + C
   3 = Strong use of diffusion = A + A

3. Low - High use of open confrontation and conflict.

   1 = No use of open conflict = A + A or C + C or A + C.
   2 = Some use of open conflict = A + B or B + C.
   3 = Strong use of open conflict = B + B.

SCORING Enmeshment /Overprotection

1. Sum the scores for problems 1 and 2 on the following concepts:

   Mind reading
   Mediating response
   Alliance shifts
   Closeness
   Overprotection

   The scores can thus range from 0 to 20: ie from no evidence to very strong evidence of enmeshment/ overprotection.

   Scores 0-1 suggest no evidence of enmeshment /overprotection.
   Scores 2 to 5 suggest at least two clear examples of enmeshment/ overprotection.
   Score 6+ suggests clear evidence (at least 3 examples) of enmeshment/ overprotection.

2. It is not possible to score a problem discussion on alliance shifts and mediating response when less than three people take part in the discussion. If this happens, then pro-rate the scores.

SCORING Weak Generation Boundaries

Sum the scores from problems 1 and 2 on generation boundaries.

The scores can thus range from 0 to 4.

Score 0-1 no evidence of weak boundaries
Score 2+ evidence of weak boundaries

APPENDIX IV: LIFE EVENTS IN PAST SIX MONTHS

Instructions

To be completed by **ONE PARENT** with the held of the other parent if wished.

Overleaf you will find a list of events or situations that sometimes affect families. For each event you are asked:

Has this happened to anyone in your family circle in the past six months?

If your answer is Yes, then you are also asked to estimate how much of a problem this has been for your family on a scale from 0-4.

0 = No problem
1
2 = Some problem
3
4 = A major problem

EXAMPLE

	Yes/No	If yes 0-4
Q.12 Had trouble with relatives	Y	3

This answer indicates that, Yes, there has been trouble with relatives and that it has been a considerable problem for the family.

	Yes/No	If yes 0-4
Q.7 Got engaged or married	N	-

This answer indicates that no one got engaged or married in the last six months and therefore the "problem" question does not apply.

Notes

"Family" here refers to anyone living in your household or any relative that you are very close to.

Sometimes two questions could refer to the same event. For example

Q.10 Had a baby
Q.11 New person joined the household

If a baby has been born and joined your household, give only one "Yes". You would answer "Yes" to both Q.10 and Q.11 if, for example, a baby was born and a lodger joined your household.

LIFE EVENTS IN PAST SIX MONTHS

Has this happened to anyone in your family circle in the past six months?	Yes/No	If yes 0-4
1. Retired from work		
2. Started or left a job		
3. Started or left school or college		
4. Had trouble at work		
5. Had a major change in work conditions or responsibilities		
6. Sat important exams		
7. Got engaged or married		
8. Family member left home		
9. Became pregnant		
10. Had a baby		
11. New person joined the household		
12. Had trouble with relatives (outside household)		
13. Death of person in household		
14. Death of other close family member or friend		
15. Major accident or injury		
16. Major change in health of family member (other than child in research study)		
17. Major change in quality of marital relationship		
18. Divorce or break up of special relationship		
19. Major change in financial situation		
20. Loss, damage or theft of property		
21. Had trouble with the police or law		
22. Moved house or change in housing conditions		

APPENDIX V: EDINBURGH FAMILY SCALE

On the following pages you will find some statements about your family as a whole. Please read each statement and decide how well the statement describes your family. Put a tick in the box that best describes your family.

	Strongly agree	Agree	Disagree	Strongly disagree	For office use only E R C
Example: We disagree on lots of things		✓			
1. When somebody in our house gets hurt or upset, we all react					( )
2. We like to smooth things over					( )
3. We spend too much time arguing about what our problems are					( )
4. Other people's suggestions about our family tend to be rather a waste of time					( )
5. When problems come up, we try different ways of solving them					( )
6. We prefer doing things at home to going out on our own					( )
7. We never let things pile up until they are more than we can handle					( )
8. I feel responsible for family members					( )
9. We see no need to change our way of doing things					( )
10. We are very concerned about each other					( )
11. When things aren't going well, it takes too long to work them out					( )
12. Old ways of doing things tend to be the best					( )

		Strongly agree	Agree	Disagree	Strongly disagree	For office use only E R C
13.	It's better to go along with what other people say in the family					( )
14.	Parents are always around for the children					( )
15.	We deal with our problems even when they are serious					( )
16.	My parents/children are just like close friends					( )
17.	We prefer things to stay the same in our family					( )
18.	We have disagreements that can't be talked about					( )
19.	Our way of life does not need to change					( )
20.	The least thing can cause an upset in our family					( )
21.	Family ties are more important to us than friendships					( )
22.	We never seem to get to the bottom of family problems					( )
23.	When one family member has a problem, everyone worries about him or her					( )
24.	In our family we like things to be cut and dried					( )
25.	Family members are very involved in each other's lives					( )
26.	The same old arguments come up again and again					( )
27.	Rules change in our family					( )

Scoring Guide

1. There are three subscales:

Enmeshment/ Overprotection	Qs 1, 6, 8, 10, 14, 16, 21, 23, 25
Rigidity	Qs 2, 4, 9, 12, 13, 17, 19, 24, 27
Conflict	Qs 3, 5, 7, 11, 15, 18, 20, 22, 26

2. Place scoring template over questionnaire and put score in appropriate brackets. Scores range from 3 to 0. The higher the score the more dysfunctional the family.

3. Sum the scores for each subscale.

4. Refer to the Transformation tables. There is one table for adults (parents) and one for adolescents (children). Transform the raw score total for each subscale to a standard score. For example: If an adult family member obtained a raw score of 15 on the Enmeshment (E) subscale, this would translate from the table to a standard score of 42. A raw score of 22 on the Rigidity (R) subscale would translate from the table to a standard score of 81.

Norms for Edinburgh Family Scale (ADULTS)
Transformed standard scores with a mean of 50 and SD of 10

Raw Score	Enmeshment/ Over-protection	Rigidity	Conflict	Raw Score
0				0
1			21	1
2			22	2
3			28	3
4			32	4
5		21	35	5
6		25	38	6
7		28	42	7
8		31	45	8
9		35	49	9
10		39	52	10
11	27	42	55	11
12	31	46	59	12
13	35	50	62	13
14	38	53	65	14
15	42	56	68	15
16	45	60	72	16
17	49	63	76	17
18	53	67	79	18
19	56	70	82	19
20	60	74		20
21	63	77		21
22	67	81		22
23	71	84		23
24	74	87		24
25	78			25
26				26
27				27

Norms for Edinburgh Family Scale (ADOLESCENTS)
Transformed standard scores with a mean of 50 and SD of 10

Raw Score	Enmeshment/ Over-protection	Rigidity	Conflict	Raw Score
0				0
1				1
2				2
3			27	3
4			30	4
5		22	33	5
6		26	35	6
7	27	29	38	7
8	29	32	41	8
9	32	35	43	9
10	35	39	46	10
11	38	42	49	11
12	41	45	52	12
13	44	49	54	13
14	47	52	57	14
15	50	56	60	15
16	53	58	62	16
17	56	62	65	17
18	59	65	68	18
19	62	68	70	19
20	65	72	73	20
21	67	75	75	21
22	70		78	22
23	73		81	23
24	76		84	24
25	79		86	25
26			89	26
27				27

APPENDIX VI: STEIN AND RIESSMAN SCALE[1]

Each parent living in the household will need to complete a copy of this scale.

Name: .....

Here are some statements that people have made about living with a young person who is ill. For each statement you are asked to say whether you strongly agree, agree, disagree or strongly disagree with it.

Please put a tick in the box that best describes your situation as it is now.

	Strongly agree	Agree	Disagree	Strongly disagree	For office use only F S P M
Example: My relatives interfere and think they know what is best for my child			✓		
1. The illness is causing financial problems for the family					( )
2. Our family gives up things because of my child's illness					( )
3. People in the neighbourhood treat us specially because of my child's illness					( )
4. Not many people understand the burden I carry					( )
5. Time is lost from work because of hospital appointments					( )
6. We see family and friends less because of the illness					( )
7. I don't have much time left over for other family members after caring for my child					( )
8. I/my partner is cutting down the hours I work to care for my child					( )
9. Travelling to the hospital is a strain on me					( )

		Strongly agree	Agree	Disagree	Strongly disagree	For office use only F S P M
10.	Learning to manage my child's illness has made me feel better about myself					( )
11.	We have little desire to go out because of my child's illness					( )
12.	Sometimes I feel as if we live on a roller coaster: in crisis when my child is very unwell, OK when things are stable					( )
13.	Because of what we have shared, we are a closer family					( )
14.	I live from day to day and don't plan for the future					( )
15.	Because of the illness, we are not able to travel away from home					( )
16.	Sometimes we have to change plans about going out at the last minute because of my child's state					( )
17.	Fatigue is a problem for me because of my child's illness					( )
18.	My partner and I discuss my child's problems together					( )
19.	My relatives have been understanding and helpful with my child					( )
20.	I think about not having more children because of the illness					( )
21.	Additional income is needed in order to cover expenses of the illness					( )
22.	We try to treat my child as if he/she were a normal child					( )
23.	I worry about what will happen to my child in the future					( )

Scoring

1. Scores can range from 1 to 4.

2. Use the template to obtain transformed scores. The scores for each of the four factors, Financial, Familial/Social, Personal Strain and Mastery, are obtained by simply summing the items on that particular subscale.

Factors:

Financial	Items 1 + 5 + 8 + 21 (4)
Familial/Social Items	Items 2 + 3 + 6 + 7 + 11 + 15 + 16 + 20 (8)
Personal Strain	Items 4 + 9 + 12 + 14 + 17 + 19 + 23 (7)
Mastery	Items 10 + 13 + 18 + 22 (4)

High score = high impact or poor coping

[1] Stein,R.E.K., & Riessman,C.K. (1980) The Development of an Impact-on-Family Scale. Preliminary findings. Medical Care, 15, 465-472.

APPENDIX VII: FAMILY ASSESSMENT MEASURE (Skinner et al, 1983)

Each member of your household 13 years and older will need to complete a copy of this scale.

Name: .....

On the following pages you will find some statements about your family as a whole. Please read each statement and decide how well the statement describes your family. Put a tick in the box that best describes your family.

	Strongly agree	Agree	Disagree	Strongly disagree
Example: We disagree on lots of things		✓		
1. We spend too much time arguing about what our problems are				
2. Family duties are fairly shared				
3. When I ask someone to explain what they mean, I get a straight answer				
4. We are as well adjusted as any family could possibly be				
5. When problems come up, we try different ways of solving them				
6. My family expects me to do more than my share				
7. We argue about who said what in our family				
8. My family could be happier than it is				
9. I don't see how any family could get along better than ours				
10. We never let things pile up until they are more than we can handle				
11. We agree about who should do what in our family				
12. I never know what's going on in our family				
13. My family and I understand each other completely				

		Strongly agree	Agree	Disagree	Strongly disagree
14.	When things aren't going well, it takes too long to work them out				
15.	We can't rely on family members to do their part				
16.	We take the time to listen to each other				
17.	Some things about my family don't entirely please me				
18.	We deal with our problems even when they are serious				
19.	One family member always tries to be the centre of attention				
20.	My family lets me have my say, even if they disagree				
21.	My family is not a perfect success				
22.	I don't think any family could possibly be happier than mine				
23.	When someone in our family is upset, we don't know if they are angry, sad, scared or what				
24.	You don't get a chance to be an individual in our family				
25.	When I ask why we have certain rules, I don't get a good answer				
26.	We have the same views on what is right and wrong				
27.	Some days we are more easily annoyed than on others				
28.	We tell each other about things that bother us				
29.	We feel loved in our family				
30.	When you do something wrong in our family, you don't know what to expect				
31.	It's hard to tell what the rules are in our family				
32.	Sometimes we are unfair to each other				

		Strongly agree	Agree	Disagree	Strongly disagree
33.	I can let my family know what is bothering me				
34.	We never get angry in our family				
35.	My family tries to run my life				
36.	If we do something wrong, we don't get a chance to explain				
37.	We argue about how much freedom we should have to make our own decisions				
38.	We sometimes hurt each other's feelings				
39.	When someone is upset, we don't find out until much later				
40.	Sometimes we avoid each other				
41.	We feel close to each other				
42.	Punishments are fair in our family				
43.	The rules in our family don't make sense				
44.	We never get upset with each other				
45.	When our family gets upset, we take too long to get over it				
46.	We always admit our mistakes without trying to hide anything				
47.	We don't really trust each other				
48.	We hardly ever do what is expected of us without being told				
49.	We are free to say what we think in our family				
50.	We have never let down another family member in any way				

Scoring

There are 7 clinical subscales plus subscales for Social Desirability and Denial.

Task Accomplishment	Items 1, 5, 10, 14, 18
Role Performance	Items 2, 6, 11, 15, 19
Communication	Items 3, 7, 12, 16, 20
Affective Expression	Items 23, 28, 33, 39, 45
Affective Involvement	Items 24, 29, 35, 41, 47
Control	Items 25, 30, 36, 42, 48
Value and Norms	Items 26, 31, 37, 43, 49
Social Desirability	Items 4, 8, 9, 13, 17, 21, 22
Denial	Items 27, 32, 34, 38, 40, 44, 46, 50

1. Scores for each item range from 3 to 0. The higher the score, the more dysfunctional the family.

2. The scores are summed for each subscale and then converted to standard scores using conversion tables.

Obtainable from:

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APPENDIX VIII: COOPERMAN SCALE (Cooperman et al, 1971)

2	1	0
Activity 1. Engages in athletics with normal peers 2. Full activity	Attends regular school with normal peers - misses not more than 2 days a month	Misses more than 2 days per month and no physical activity
Chest x-ray Normal	Minimally increased markings and emphysema	Well defined changes with emphysema, marked streaking and nodularities
Clubbing 0 to 1+	1+ to 2+ Definite clubbing with no cyanosis of nail beds	2+ and greater
Growth and Development Above 25th percentile for height and weight	Above 3rd percentile for height and weight	Below 3rd percentile for height and weight
Complications None	Minimal to moderate emphysema and/or widespread focal crepitations and rales	Cirrhosis of liver and portal hypertension, cor pulmonale and widespread bronchiectasis with advanced emphysema, or persisting widespread rales and crepitations